The research priorities for UK ENT, Hearing and Balance Care:

A qualitative investigation of the views and voices of stakeholders

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Declaration

I, Natalie Diane Bohm, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Acknowledgements

Abstract

Clinical research activity in ENT, Hearing and Balance care has historically been low in the UK and internationally. Stimulating this activity can improve patient care but it has been recognised that there is significant waste in biomedical research, and that research questions need to be developed that translate to patient benefit. Priority setting has been increasingly used to develop research agendas to address this, reduce waste, and influence the allocation of resources in healthcare research. Although there have been previous research priority setting exercises in ENT, Hearing and Balance care, none undertaken in the UK have gone beyond a specific clinical topic or consulted with a broad range of stakeholders.

This thesis, as the first study of the research priorities across UK ENT, Hearing and Balance care, contributes to addressing this gap. Following a review of the literature a multi-step process using qualitative methodology was designed to determine stakeholders and identify research ideas. Gathering data through a survey and focus groups, developing the ideas and then prioritising them through an Expert Forum and Consensus Conference, this study addressed the question: **What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?**

A two-dimensional framework, inductively derived from the data, identified research themes and clinically-based topics. Within this framework, 21 research priorities were chosen by mixed groups of stakeholders through facilitated small and large group discussion, and reasons given by the groups for their choices were outlined. This study discusses the quality of the process developed, and the priorities

compared to those from other exercises. In developing the priorities, the balance

needed between methodological purity and pragmatism is considered, and the

contribution of this study to research in the field is explored.

Impact Statement

Biomedical research consumes significant resources, and these may be wasted through a poor choice of research questions. This research is important, both to improve clinical outcomes and to inform allocation decisions for health service design. There have been concerns about the volume and quality of clinical research being done in the fields of ENT, Hearing and Balance care. In the UK this lack was seen to have an impact on services, with growing concerns that rationing was occurring due to a perceived lack of evidence. This resulted in a recognition of the need for coordinated and aligned research efforts in these fields. It was felt that this would stimulate future research funding and activity which would form the evidence base for developing and maintaining services. In order to use research resources effectively, it was determined that a research agenda was needed.

This thesis is the academic study on which the UK ENT, Hearing and Balance care research agenda was based. Its aim was to determine the research priorities for UK ENT, Hearing and Balance care according to the views and voices of its stakeholders. No previous multi-stakeholder study of these priorities had been done in the UK. The findings of this thesis, a framework for UK ENT, Hearing and Balance care research priorities and the 21 priorities inductively developed from stakeholder responses, have been used to stimulate research and funding in the field. Research networks have also developed, initiated by the relationships forged in the exercise and the example of collaboration it provided. These are considered in detail in the final chapter of this thesis. The impact of the research developed from the priorities on outcomes and services is a field for further study.

This thesis has also made a methodological contribution to research priority setting by using a qualitative approach with a variety of methods and framework analysis for the first time. Research priority setting is an evolving field, and one of the concerns is that the methods used do not always optimise inclusion, especially for marginalised groups, and that the original meanings of stakeholder responses may be lost or changed. The approach used here allowed the incorporation of the views of a range of stakeholders and developed a shared understanding of priorities. This provides an example for those designing research priority setting exercises in the future, offering a way to maximise inclusion, with the potential to impact on future equitable priority setting.

Table of Contents

Index of Tables

Index of Figures

List of Abbreviations

AAP   An ageing population (small group in Consensus Conference)

AHLT   Adult hearing loss and tinnitus (small group in Consensus Conference)

AoHL   Action on Hearing Loss

AOM   Acute otitis media

BAA   British Academy of Audiology

BP   Balance problems (small group in Consensus Conference)

BPPV   Benign paroxysmal positional vertigo

BRC   Biomedical Research Centre

BSA   British Society of Audiology

CBT   Cognitive behavioural therapy

CCG   Clinical commissioning group

COHRED   Council on Health Research for Development

CYP   Children and young people (small group in Consensus Conference)

ENT   Ear, Nose and Throat Surgery

ENT UK   The British Association of Otorhinolaryngology - Head and Neck Surgery

evidENT   UCL Centre for evidence-based ENT

GAfREC   Governance Arrangements for Research and Ethics Committees

GPs   General practitioners

HCP   Health care professional

HMIC   Health Management Information Consortium

HNA   Head, neck and airway (small group in Consensus Conference)

IIA   Inflammation, infection and allergy (small group in Consensus Conference)

Ix   Investigations

JLA   James Lind Alliance

HRA   Health Research Authority

MeSH   Medical Subject Headings thesaurus

NHS	National Health Service
NHS R&D	NHS Research and Development Office
NIHR	National Institute of Health Research
NIHR CCRN	NIHR Comprehensive Clinical Research Network
NRES	NHS Research Ethics Service
OSA	Obstructive sleep apnoea
PICO	Population, intervention, comparator, outcome
PPI	Patient and public involvement
PSP	Priority Setting Partnership
RCP	Royal College of Physicians
RCS (Eng)	Royal College of Surgeons of England
RCT	Randomised controlled trial
REC	Research ethics committee
RNTNE	Royal National Throat, Nose and Ear Hospital
RPSE	Research priority setting exercise
SLT	Speech and Language Therapy/Therapist
UCL	University College London
UCLH	University College London Hospital
UK	The United Kingdom
VEMP	Vestibular evoked myogenic potentials
vHIT	Video head impulse test

Chapter 1: Introduction

Exercises to determine research priorities have become more common both internationally and in the UK. They have been used to develop research agendas to influence the allocation of funding for healthcare research, and to try to reduce their waste. As they aim to influence allocation, exercises in priority setting can be considered innately political acts.

Setting priorities for research also has philosophical, ethical and social dimensions as it involves making value judgements, choosing between competing projects and ideas for limited resources. This means that the process of choosing these priorities is complex and can have an impact on a range of different stakeholders.

Multiple processes using different methodologies and methods have been used to set research priorities, and there is no consensus as to the best approach. Some of these use strictly technical approaches based on health economics and burden of disease. Others seek to identify and engage a range of stakeholders using social processes. Some seek to combine both these approaches.

The heterogeneous nature of approaches and processes used to develop priorities and research agendas may appear to show a lack of consensus. However, this is viewed as both appropriate and inevitable (Viergever *et al* 2010), and what is important is getting the process right for the setting of the exercise (Lomas *et al*

2003). In setting priorities for health care this has been seen as crucially important in ensuring that they are "socially acceptable" (Klein *et al* 1999).

This thesis outlines the design and findings of a research priority setting exercise for ENT, Hearing and Balance care in the UK. It addresses the need for these priorities to come from the views of a range of stakeholders. This chapter provides the background for this thesis. It discusses why there was a need for a research agenda for ENT, Hearing and Balance care and the state of research prior to this exercise. It outlines research priority setting in general and discusses the diversity of priority setting. Finally, it outlines the structure for the rest of the thesis.

1.1 The origins of this thesis

In 2013 Søreide *et al* stated that despite over 235 million patients undergoing surgery worldwide each year, less than 1% were enrolled in surgical trials.
It was recognised that there were evidence gaps in basic surgical care, and that surgery particularly seemed to have a disproportionately large number of unanswered fundamental clinical questions (The Lancet 2013). This was recognised as an issue for commissioners of surgical services who "want to be confident of receiving consistent high-quality care based on the best available evidence" (The Lancet 2013, p 1071). This had been seen as an ongoing international issue, and it was one that had an impact on the provision of services in the UK.

In 2006 Croydon Primary Care Trust in London identified 34 surgical procedures that were described as being of 'limited clinical effectiveness' (Klynmann 2010). It was

decided that they would either be decommissioned or subject to access criteria. The rationale for this was that there was no evidence of benefit to all the patients undergoing these procedures. Furthermore, it was noted that reducing or stopping them could result in cost savings. The following year, the London Health Observatory estimated the savings that would have been made if access to the 'Croydon List' procedures had been limited across London. If this had been done in 2005/2006, then the potential cost savings would have been between £28 million and £93 million (Malhotra and Jacobsen 2007).

In 2009 the UK National Health Service (NHS) was tasked with finding efficiency savings of £20 billion by 2014/15 (Nicholson 2009). As part of their economic strategy to find these savings, service commissioners refused to commit resources to procedures and pathways that they felt had insufficient evidence. As a result, over two-thirds of Primary Care Trusts restricted access to such treatments on the basis that the evidence of their effectiveness was limited (Audit Commission 2011). Many of these restrictions were based on the original 'Croydon list.' They were not based on a centralised list and were not done in consultation with wider stakeholders, including members of the public (Fawcett *et al* 2011; Klynmann 2010). A Royal College of Surgeons survey from 2014 noted the lack of consistency between the Clinical Commissioning Groups (CCGs, which had superseded the Primary Care Trusts) and the lack of a clinical basis for the restrictions (Edwards *et al* 2015).

Cost saving then extended beyond surgical procedures, to impact on the provision of other interventions and services where there had been deemed to be insufficient evidence of benefit. For example, the North Staffordshire CCG proposed restricting

access to NHS hearing aids, mainly for elderly people with mild hearing loss, and increasing the threshold of eligibility for those with moderate hearing loss. The CCG explicitly stated that they did not accept that the evidence supported the continued funding of these aids (North Staffordshire Clinical Commissioning Group 2015, Enclosure 11).

Although these restrictions appeared to be confined to England, there were indications that the devolved nations were affected by this move to restrict services in the name of efficiency. Action on Hearing Loss produced two reports on service cuts. The 2012 report (Calton 2012) noted that there were increasing restrictions in provision of some audiology interventions and services in Wales, as well as England. The second report (Lowe 2015) gathered information from all four devolved nations. It showed a failure to invest in services across the UK, with a third of audiology providers having to reduce their services over the previous two years.

Ear, Nose and Throat (ENT), Hearing and Balance services were felt to be under particular threat of rationing throughout the UK during this time. The original 'Croydon list' of procedures included several ENT operations, including sinus surgery, tonsillectomy and ventilation tube (grommet) insertion in the ear drum. Audiology services, as seen above, were also felt to be at risk. ENT UK, the British Society of Audiology (BSA) and the British Academy of Audiology (BAA), the main professional bodies in these specialties, felt that the threats to multiple services warranted urgent attention. During the immediate discussions with policymakers about how services should be provided going forward, it was recognised that longer-term strategies were needed to assess the 'value' of interventions and services.

The Federation of Surgical Specialty Associations had expressed its concerns that the evidence for developing the lists restricting procedures was poor, and that as a result these differed between Clinical Commissioning Groups. The Association expressed its commitment to working with policymakers to provide clinical leadership to ensure high quality and cost-effective healthcare (Fawcett *et al* 2011). Part of the Action on Hearing Loss strategy in their 2015 report (Lowe 2015) was to affirm their commitment to produce and share research findings to help providers and commissioners improve the quality of audiology services.

The professional bodies supported the need for further coordinated and aligned research efforts. It was felt that this would stimulate future research funding and activity which would form the evidence base for developing and maintaining services. It was also felt that these efforts would raise the national profile of the specialties with policymakers, the public and the press. The organisations hoped that this increased profile would improve access to negotiations with service commissioners, demonstrate their commitment to maintaining patient care to their memberships and ensure that services were fit for purpose. Clearly there were many drivers behind cutting funding to interventions and services, however lack of evidence was identified as key. The relatively low levels of research activity and resultant low level of high-quality clinical evidence was recognised to have had a practical impact on the clinical practice of the specialties in the UK.

At the same time that these service restrictions were being considered, the clinical research community in England was undergoing a transformation. In 2006 the

National Institute of Health Research (NIHR) was established. It aimed to provide a framework to support research, researchers and infrastructure in the NHS in England (NIHR 2013). ENT, Hearing and Balance care benefitted from this focus on clinical research, with the appointment of an NIHR Research Professor in 2011 whose remit was to address the deficit of high-quality clinical research in these fields.

This appointment stimulated discussions between ENT UK, the BSA, the BAA and the clinical academic community. There was shared concern and frustration, especially in considering some areas where commissioners felt there wasn't sufficient evidence of benefit, such as the use of grommets in children or the efficacy of tonsillectomy. Here the clinical and academic communities felt that these questions had been or were in the process of being addressed. For example, the Scottish Intercollegiate Guidelines Network (SIGN) guideline for management of sore throat (SIGN 2010) considered the evidence for surgery for children with recurrent tonsillitis. This aspect of the guideline was based on clinical trial evidence (Paradise *et al* 1984) and was widely accepted as a basis for UK clinical practice. Thus, stopping all tonsillectomies was seen as going against evidence-based practice. Through these discussions it was recognised that there was a need to produce high quality, high value evidence addressing a variety of perspectives: the needs of patients, the concerns of policymakers, and to underpin the clinical practice of the specialties. These discussions resulted in financial and professional support for a research priority setting exercise in ENT, Hearing and Balance care, which became known as GENERATE.

In order to fulfil the aims of the professional bodies and clinical research community, to justify the investment of these communities, and to develop evidence that would have clinical impact and shape policy, it was recognised that this exercise would need to be rigorous. It was agreed that an academic approach would be used to accomplish this. The development of this approach, the methodology used, and the analysis and findings of the exercise form the basis of this thesis.

To refine the research question for this thesis discussions were had with people who had run their own exercises. Exercises that were running in other specialties were also observed. Background reading prior to the formal literature review was also done. These activities, and the discussions with professional bodies, gave insight into research priority setting and why it has been done, as well as the current state of clinical research for ENT, Hearing and Balance care, which form the basis of the following sections.

1.2 Why do we need priority setting in research?

Globally, biomedical research consumes significant financial resources (Macleod *et al* 2014). It was estimated that approximately 85% of this investment may have been wasted through avoidable causes (Chalmers and Glasziou 2009). This waste was seen to be multifactorial, but one of the key areas highlighted was waste through a poor choice of research questions, resulting in low priority questions being chosen and important outcomes not being assessed (Chalmers and Glasziou 2009).

Other concerns have been expressed about the use of biomedical research resources. It was recognised that this research has been highly valued by the public, governments and industry. However, the global financial recession of 2007 – 2009 led to increased focus on allocation of public and private research expenditure (Dorsey *et al* 2010). This scrutiny led to growing questions as to whether the investment in biomedical research, where there was increased understanding of the mechanism of disease, was translating into clinical benefit. This divide between basic and clinical research was felt to require a reset in research culture with greater collaboration between multidisciplinary groups (Butler 2008).

As a result of this perception of wasted resource and potential lack of benefit to patients, increasing focus was placed on who set the research agenda, with concern that the vested interests of industry and career researchers had been allowed to dominate (Greenhalgh *et al* 2014; Oliver *et al* 2004). Involving patients and clinicians in setting priorities was suggested as a way of redressing this concern, reducing the waste of research investment, increasing the value of research agendas and providing greater clinical impact (Chalmers and Glasziou 2009; Oliver *et al* 2004; Butler 2008).

This global concern was considered in the context of the UK. The UK Health Research Analysis 2014, conducted by the UK Clinical Research Collaboration (2015), analysed the health research landscape between 2004 and 2014. This estimated that a total of £8.5 billion was spent on health research and development in the UK in 2014. This represented a real term decrease from the estimate for the spend in 2009/10, due to a decrease in pharmaceutical spend in this area. The

analysis focused on public and charitable health-relevant research in the UK. It showed that public funding (both via Government funding and donation via medical research charities) increased through that decade. However, this would appear to have occurred between 2004 and 2009; between 2009 and 2014 total funding in real terms flattened across public and charity sectors, with a compound annual growth rate of 1.4% during this time. To put this into wider context, UK investment in public-funded research dropped below 0.5% of GDP in 2012, putting UK government support for science funding last in the G8 group of countries (Rohn, Curry and Steele 2015).

This was important context for GENERATE. When this research priority setting exercise was being planned, funding for health research in the UK was under pressure with public levels of funding flattening or decreasing. Competition for these resources was seen to be likely to increase, with increased demands to show that future research would provide value. As stated above, one way to demonstrate this potential value was to establish research priorities and set research agendas using the key stakeholders identified by Chalmers and Glasziou (2009) and Oliver *et al* (2004).

Resources to conduct health research extend beyond the funding required for projects. Clinicians as well as academic scientists are needed for health research as their regular clinical contact allows them to identify key health needs and appreciate what is translatable into practice (Royal College of Physicians (RCP) 2016). Although research funding can be allocated to clinicians for their time, the demands of their clinical practice present a challenge. In a survey of 2000 UK doctors, the

RCP (2016) found that time, as well as funding, was one of the biggest barriers to doctors doing more research. Again, it was recognised that in order to maximise the use of this limited resource priorities for research needed to be set.

How to set these priorities became the next question. In order to establish this, greater understanding of how priority setting has been used and different ways of setting them was required.

1.3 The diversity of priority setting

Priority setting has been used in diverse disciplines. Within healthcare it was traditionally used in service delivery and policy development (Campbell 2010). It was then increasingly used in setting priorities for healthcare research. Following the launch of the NHS Research and Development programme in 1991, priority setting in healthcare research became more coordinated and increasingly systematic (Oliver *et al* 2004). The adoption and adaptation of processes and methods from other sectors and settings was inevitable, with the lessons learned from other areas being translated into priority setting in this field (Lomas *et al* 2003).

Lomas *et al* (2003) identified two broad approaches to priority setting: technical assessments and interpretive assessments. The former relies on quantifiable disease-based data. An example of this is the Burden of Disease Approach, where the magnitude and determinants of disease are analysed, along with the current knowledge base and cost-effectiveness of, and resources available for, research in order to determine priorities (Montorzi *et al* (2010)). Griffiths *et al* (2002) noted that a

purely disease-based data method limits the approach as it does not consider criteria

beyond disease burden and costs. Furthermore, it is not easily applicable to other

dimensions of research, such as setting or methodology. Other limitations that have

been noted are that these approaches may not reflect the values of the broader

population (Lomas *et al* 2003); these methods tend not to be used as they are

resource-intensive (Gagliardi *et al* 2008); and are of limited use where there is little

data available (Montorzi *et al* 2010).

Interpretive exercises may utilise technical data within a broader context, but at their

heart they rely on the subjective judgments of participants expressed through

structured exercises (Lomas *et al* 2003). They have the added advantage of allowing

stakeholders to question the assumptions on which technical data is based (Lomas

et al 2003). This means that there is the possibility to question the universal

acceptance of scientific rationale and method by the stakeholders, though this is

often not adopted in the exercises (Campbell 2010).

Exercises using an interpretive approach recognise the social nature of priority

setting exercises, based on developing dialogue, relationships and trust, where

arguably the process is more important than the end product of a list of priorities

(Abma and Broerse 2010; Campbell 2010). This approach allows one to:

"…increasingly treat knowledge as a multi-faceted public good: demanded, shaped

and used by the many stakeholders it has always claimed to serve" (Campbell 2010

p 55).

When the professional organisations and researchers considered the aim of GENERATE, they recognised that a wide range of stakeholders would need to be consulted. The negative response from the public when the 'Croydon list' and the various CCG restrictions were developed without wider consultation was a lesson: ignore stakeholders at your peril where resource decisions are being made in healthcare. It was also recognised that ENT, Hearing and Balance conditions are managed across different specialties and settings. Thus, any attempt to increase research activity and adopt the resulting evidence would require the engagement of multiple different stakeholders and potentially the development of national collaborations.

Based on this rationale, interpretive assessments were selected as the most appropriate for a multi-stakeholder priority setting exercise. The focus on the social nature of the exercise, and the ability to challenge established assumptions was felt to be more appropriate where a range of stakeholders could have a variety of views and values. This informed the scope of the literature review in the second chapter, which is outlined further on in this chapter.

1.4 ENT, Hearing and Balance care research: The Global perspective

As well as global concerns about the levels of waste in biomedical research, there were specific concerns about the volume and quality of clinical research being done in the fields of ENT, Hearing and Balance care. Traditionally, the number of publications in discovery science in ENT, Hearing and Balance care have been comparable or better to similar surgical specialties (Bhattacharyya 1999), although

there were concerns that scholarly output had been declining over the last 30 years (Bhattacharyya and Shapiro 2007). However, doubts had been raised about whether this was translating into clinical research.

Sharp, Harvey and Burton (2007) conducted an evaluation of the global knowledge base for ENT, Hearing and Balance care. This study identified the number of ENT and audiology publications in MEDLINE based on MeSH headings for conditions. This number was compared to the total number of records in the database. They found that publications for ENT and audiology conditions accounted for 1.5% of the entire database. The authors noted that this did not reflect the frequency of consultation for these conditions in primary care. They also analysed the number of publications based on the classical evidence levels from systematic reviews to case reports. They found that the majority of these publications were of what is traditionally considered a lower level of evidence, with case reports being the most frequently published. Systematic reviews and randomised controlled trials comprised less than 1% and 5% of prevalent knowledge respectively in the majority of conditions. This study concluded that there was a need to seek "better evidence on which to base current practice" and that this process should be open to patients, policy makers and health resource managers.

A more recent analysis of clinical trials activity for ENT in America from 2007 – 2010 came to similar conclusions with regard to the need to increase clinical research in the specialty (Witsell *et al* 2013). This analysis of registered ENT and Head and Neck Surgery clinical trials in the government clinical trials database showed that less than 3% of interventional trials registered were assigned as ENT-related

research, with health services the least common area of study. This research activity was compared to data on clinical visit volumes to a large tertiary medical centre for ENT-related diagnoses, which showed poor correlation with ENT disease prevalence. The study authors concluded that there was a need for increased research activity to complement the relative prevalence of these conditions, but recognised the challenge presented by the heterogeneity of the specialty and the fact many patients with ENT diagnoses were seen by non-ENT specialists. They felt that there was a need for a collective focus on research initiatives, with studies performed through practice-based research networks.

These studies both indicated the need for increased clinical research activity in ENT, Hearing and Balance care. They called for focused, coordinated efforts involving a range of healthcare professionals involved in treating patients with "ENT" diagnoses, as well as other key stakeholders that develop, use and benefit from research. These papers did not propose how this should be accomplished and did not specifically advocate setting research priorities as a means to achieving these ends. However, the use of interpretive priority setting methodology that fosters dialogue and relationships between these stakeholders was felt to build the basis for the necessary coordination and collaboration to develop research, especially where resources were limited.

1.5 ENT, Hearing and Balance research activity in the UK

The previous section considered research in ENT, Hearing and Balance care from an international perspective. This section discusses the state of research activity in the UK at the time that GENERATE was being initiated.

In 2002, Fenton *et al* identified the articles in ENT, Hearing and Balance journals that had achieved "classic citation status" over the twentieth century. This was defined as articles that had been cited 100 times or more. This status was considered to be a surrogate measure for the quality of the research due to the impact that it had made on the relevant scientific community. In this paper, out of 80 articles identified as citation classics, 67 originated in American institutions. Two came from the UK. Although there were confounding factors to this overview, such as the tendency of American authors to cite American articles, this insight into seminal advances in ENT, Hearing and Balance research did not highlight the UK as a leader in research in these fields.

Clinical research activity in ENT, Hearing and Balance care has historically been low in the UK compared with other specialties. This was evidenced by the low number of clinical studies in 2013 adopted onto the National Institute of Health Research (NIHR) Specialty Group research portfolio (NIHR CCRN Specialty Group Performance Management Report for June 2013: see Figure 1). Here studies in ENT were in the lower third of activity compared to other specialties. It was likely that other research activity existed, in the form of smaller and local studies. However, nationally recognised, rigorous cohort studies and interventional trials would appear to have been scarce.

Specialty Group Performance Management Report

June 2013

Studies and recruitment are counted once against Main Specialty Group.

Figure 1 Number of Open Studies by NIHR Specialty Group and Study Design Type (NIHR CCRN Specialty Group Performance Management Report for June 2013)

During this time, the Royal College of Surgeons published a report highlighting that research funding in surgery was a small fraction of UK government funding in medical research (Royal College of Surgeons of England 2011). This indicated under-investment in all research activity in surgical specialties, thus problems with funding were not a problem specific to ENT, Hearing and Balance care.

Concerns about the low volume of surgical research in the UK had been echoed by Cancer Research UK (National Cancer Research Institute 2012), which noted that this was true both for surgery within and beyond cancer. This identified a lack of research culture in surgery in the UK and recognised that capacity building and

research skill development were needed. Within the general field of cancer, it was felt that a consensus-building process from the surgical specialties to identify key research themes could provide bottom-up impetus to developing research. Thus, the solution that had been proposed by the ENT, Hearing and Balance organisations and research community to use multi-stakeholder engagement in interpretive processes appeared to be one that resonated with different groups.

The background to GENERATE, the research priority setting exercise for ENT, Hearing and Balance care in the UK, and the context of priority setting and research activity for the specialties, has been outlined in these sections. The research question for this thesis and an overview of the following chapters is presented in the next section.

1.6 Overview of the thesis

In the previous sections the need for a research priority setting exercise in UK ENT, Hearing and Balance care was outlined as well as the importance of involving a range of stakeholders. It was decided to use an interpretive rather than a technical approach in order to build consensus with this range of stakeholders.

In order to develop an exercise to fulfil these requirements, the overarching research question for this thesis was defined as:

What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?

In order to answer the research question, the objectives of this thesis were as follows:

- To examine the literature on research priority setting in order to determine the best approach for a research priority setting exercise;
- To design a research priority setting exercise for UK ENT, Hearing and Balance care;
- To execute the exercise and determine the research priorities based on the views and voices of the stakeholders.

This thesis has been structured into six chapters. Following this introduction, the second chapter examines the literature on research priority setting. It addresses three questions though a scoping review, considering what previous work has been done to determine research priorities in these fields, how priorities have been set across health research and what can be learned from the literature to design a study to answer this question. These questions are:

- What can be learned from the literature in order to design a research priority setting exercise for UK ENT, Hearing and Balance care that incorporates the views and voices of its stakeholders?
- What methodologies and methods have been used in research priority setting exercises for human health research in high income countries?
- What are their strengths and weaknesses in terms of incorporating the views and voices of stakeholders?

The third chapter outlines the methodology, process and methods used to determine the research priorities, based on the findings of the scoping review. It outlines the qualitative study design through a multi-stage process used to maximise inclusion of the stakeholders and details the analysis methods used.

Chapters 4 and 5 present the key findings of this study. They answer the research questions:

- Who are the stakeholders for UK ENT, Hearing and Balance care?
- What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?
- Why did the stakeholders prioritise those ideas?

Chapter 4 describes the stakeholder map that was developed for the exercise, the distribution of these stakeholders amongst participants in the exercise and the framework developed for the research ideas suggested by the stakeholders. Chapter 5 presents the research ideas, and illustrates their development into the final agreed priorities, while also mapping them back to the framework and stakeholders described in the previous chapter. Finally, it gives an overview of the reasons given by the stakeholders for choosing those priorities.

The final chapter of this thesis, Chapter 6, summarises the findings and outlines the impact of this study. It considers the contribution to research that it has made and reflects on its strengths and limitations.

Chapter 2: Scoping the literature: understanding how to design a research priority setting exercise

2.1 Aim and Research Questions

The previous chapter outlined the need to develop research priorities for ENT, Hearing and Balance care in the UK. The aim of this chapter is to review the literature to inform the design of a research priority setting exercise for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders.

This literature review addresses the following question:

What can be learned from the literature in order to design a research priority setting exercise for UK ENT, Hearing and Balance care that incorporates the views and voices of its stakeholders?

In order to answer this, two further questions were posed:

What methodologies and methods have been used in research priority setting exercises for human health research in high income countries?

What are their strengths and weaknesses in terms of incorporating the views and voices of stakeholders?

2.2 Methods

2.2.1 Framework for review

A scoping review of the literature with a systematic design based on Arksey and O'Malley's (2005) framework was carried out. The iterative, exploratory nature of this approach was suitable as research priority setting is an emerging area of research, and the aim of this review was to identify and examine a range of processes in a diverse body of literature (Arksey and O'Malley 2005, Gough 2012). With the Arksey and O'Malley (2005) framework the criteria used to identify, select and chart the data could iterate as studies were examined and there was increased familiarity with the research field.

2.2.2. Identifying and selecting studies

Key search terms relevant to the research questions above were identified and iteratively developed with the thesis supervisors (see Table 1 below; the full search strategy may be seen in Appendix 1, page 221). The search was run in the MEDLINE, EMBASE and HMIC databases in order to identify peer reviewed scientific papers, articles, monographs, reports, government documents and grey literature with a focus on UK health policy and management. The search period was from 1 January 2003 to 31 May 2013. This review was updated following the completion of the exercise to inform the discussion in Chapter 6 and is outlined in Appendix 2 (page 224).

Priority setting exercise	Health research	Methodology
Prioritisation	Clinical research	Process
Priorities	Health Services research	Consensus
Prioritise	Research agenda	Method
Health priorities	Research priorities	Approach
Agenda setting	Biomedical	Design
Research prioritisation	Healthcare	
	Healthcare services	

Table 1: Exploded search terms for scoping review

The search was extended through "snowballing" to identify publications about the development of research priority setting methodology, and to identify any exercises not published in peer reviewed scientific papers. This was done by:

- Reviewing the reference lists of the included papers.
- Contacting experts identified in the literature and through professional networks to provide additional publications and websites containing information about research priority setting methodology and exercises.

The inclusion and exclusion criteria for the publications identified are given in Table 2.

Criteria	Rationale
Inclusion criteria	
Clinical and health services research priority setting exercises for human subjects	Covered the scope of GENERATE
Topic or specialty-specific research priority setting exercises conducted at a national level	To inform the design of GENERATE which was performed at a national level
Exercises in high income countries (based on the list generated by the United Nations Statistics division (2013))	The politics and therefore processes of research priority setting in low- and middle-income countries were likely to have social and economic complexities that would not be relevant to GENERATE
Publications in English or Dutch language	Languages that reviewer could read or had access to translator with subject matter expertise
Exclusion criteria	
Papers not related to human health (for example, those in veterinary science)	Not relevant to GENERATE
Papers not related to research (for example, papers concerning prioritisation for health managerial resource allocation)	Beyond the scope of the research questions for this review; more suitable for a separate review
Papers in research areas not primarily involving health (for example, medical education, health workforce prioritisation, social work)	Beyond the scope of the research questions for this review; more suitable for a separate review
Technical prioritisation exercises based on quantifiable disease-based data only	Did not incorporate the multi-stakeholder views and voices central to the thesis research question
Identification of priority topics based purely on literature searches or systematic reviews	Did not incorporate the multi-stakeholder views and voices central to the thesis research question
Prioritisation based on a single personal or single team opinion	Did not use process relevant to multi-stakeholder exercises

Table 2 Inclusion and exclusion criteria for review

2.2.3 Data Extraction and synthesis

Data extraction was done using Microsoft Excel spreadsheets. With increased familiarity with the literature, the publications were divided into three groups: research priority setting methodology or overview publications; published RPSEs from healthcare fields other than ENT, Hearing and Balance; and publications for ENT, Hearing and Balance RPSEs. Data were extracted in the following fields for the included publications:

- Authorship, year of publication, objective of paper, methodology and method used, and key points as described by the authors for consideration in priority setting processes for the publications giving overviews of priority setting methodology and methods.

- Authorship, year of publication, health field scope, stakeholders, methodology and methods (as described by the authors of the paper) and prioritisation methodology references cited for the publications about the exercises not specific to ENT, Hearing and Balance care.

- Authorship, country, year of publication, scope of exercise, stakeholders, methodology and methods and outputs for ENT, Hearing and Balance priority setting exercise publications.

Examination of these led to a further table for methodology, methods used, numbers and types of stakeholders, and prioritisation criteria which was used to identify patterns and themes to inform the development of a new exercise.

Overarching themes to consider for developing a research priority setting exercise for ENT, Hearing and Balance care, methodologies, processes and methods were developed inductively.

2.3 Findings

2.3.1 Results of the search strategy

The database search produced 1267 publications after duplicates were removed. After screening by title and abstract using the inclusion and exclusion criteria, 361 full text publications were screened. A further 21 full text publications were identified from reference lists, and two from discussion with experts in the field. After full text review, 123 publications were included in the scoping review (see Figure 2).

```
┌─────────────────────────┐
│   Database Search:       │
│  (MEDLINE, EMBASE, HMIC) │
│                          │
│        1885              │
└─────────────────────────┘
              │
              │        ┌──────────────┐
              │        │  Duplicates  │
              ▼        │     618      │
                       └──────────────┘
┌──────────┐          ┌─────────────────────────────┐
│  1267    │─────────▶│ 906 publications excluded on │
│          │          │     title and abstract        │
└──────────┘          └─────────────────────────────┘
     │
     ▼
┌──────────┐          ┌─────────────────────────────┐
│   361    │─────────▶│ 259 publications excluded on │
│          │          │         full text             │
└──────────┘          └─────────────────────────────┘
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│  Selected    │
│ publications │
│   n=102      │
└──────────────┘
     │                ┌─────────────────────────────┐
     │                │   Other publications         │
     │◀───────────────│   identified through         │
     │                │   references and by          │
     │                │   discussion with experts    │
     │                │         (n=21)                │
     ▼                └─────────────────────────────┘
┌─────────────────────────┐
│ 123 publications included│
│ 113 RPSE (3 ENTHB, 110   │
│   other); 10 overviews   │
└─────────────────────────┘
```
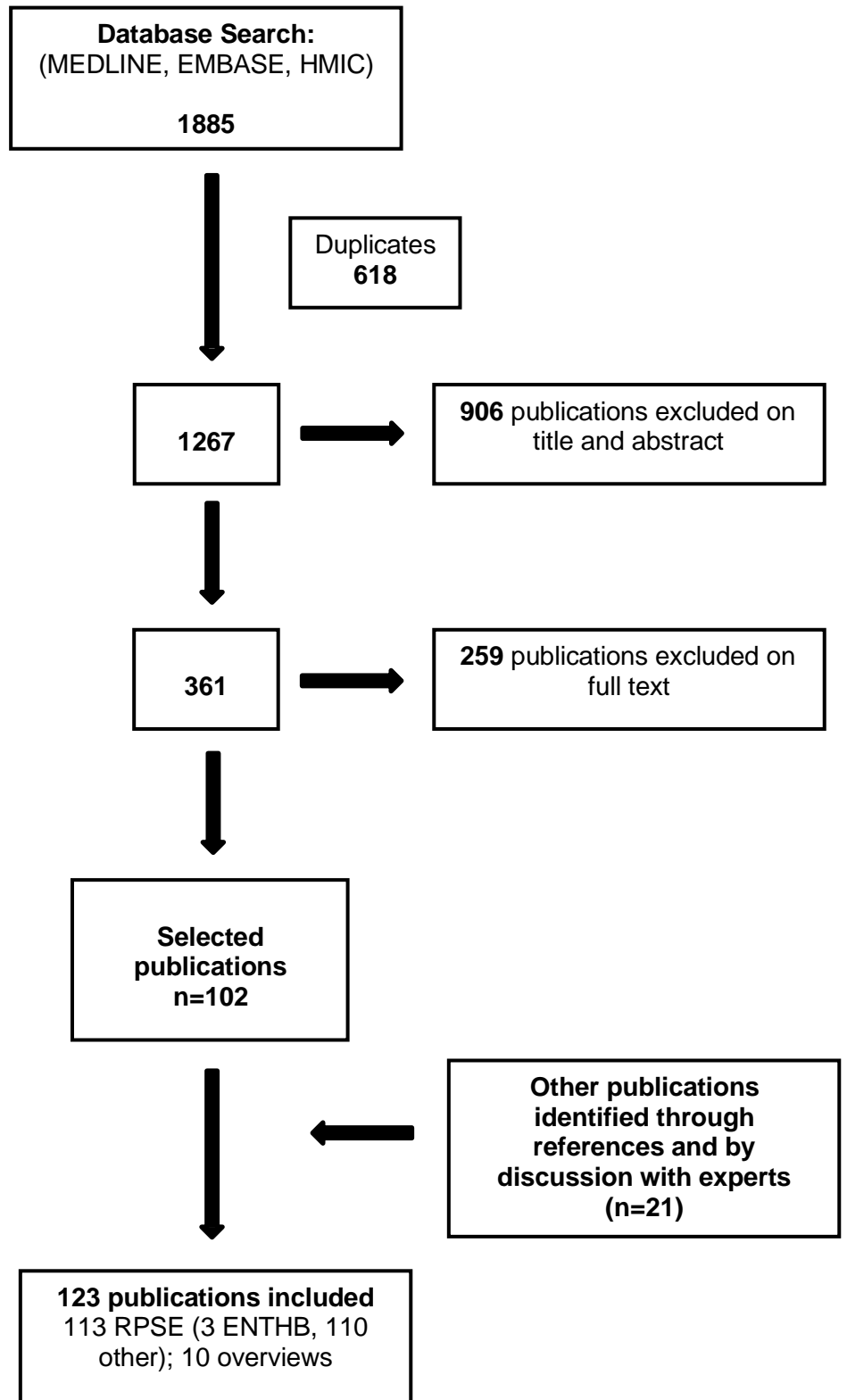
Figure 2 PRISMA flow diagram for initial scoping review

2.3.2 Designing an exercise: learning from the literature

10 publications provided an overview of methodology or best practice for research priority setting (see Table 4). They showed that there is no single priority setting process that is universally appropriate, despite continued attempts to develop this (Montorzi *et al* 2010; Viergever *et al* 2010). Neither is there a "gold standard" against which to judge the results of the exercise (Lomas *et al* 2003). Thus, the literature showed that focus has been placed on ensuring the quality of the process used to reach agreement. Common criteria on which to judge this were identified:

Transparency: This may be transparency in process and methods, the criteria used for prioritisation, or in reporting all of these. Viergever *et al* (2010) described this as involving writing a clear report about the exercise, noting who set the priorities and how they were set. Nasser, Welch *et al* (2013) stated that the need to transparently report the criteria used to set priorities was an important issue, especially if true and diverse involvement of stakeholders is to be ensured. Campbell (2010) concurred and viewed agreement for and reporting of criteria for specific exercises to be a critical step, an opinion shared by Viergever *et al* (2010). Montorzi *et al* (2010, p 13) also outlined the need for clear reporting, stating that transparency is "ensured through documentation and communication of decisions and actions, reflecting the concerns of stakeholders at national and local levels."

Inclusiveness/Fairness: Here fairness and to be seen to be fair were equated with the inclusion of relevant stakeholders. Inclusion was defined differently through some of the authors. Montorzi *et al* (2010, p 13) considered

inclusiveness to be achieved "by ensuring that all interested parties are represented throughout the process." Abma and Broerse (2010, p 171) described it as "when relevant stakeholders are enabled to participate in the process in an open and respectful way and their voice is visibly included." There is a difference in language here between representation and participation. Thus, the degree of the inclusion and the methods used to enable it become important, with Lomas *et al* (2003) requiring that these prevent domination by one group and reflect the values of all. It is important to note that this was not universally accepted, as Cowan and Oliver (2013) advocate targeted exclusion of traditional priority setters, such as academics and industry, in order to redress the historic imbalance of lack of consumer involvement.

Credibility: Results of the exercise should resonate with its stakeholders. To ensure this there was a recommendation that methods should be triangulated, and results checked with stakeholders (Abma and Broerse 2010). Credibility was also seen to be enhanced by accountability and a clear audit trail from initial gathering of ideas to final priorities (Campbell 2010; Cowan and Oliver 2013).

"Equity", as another dimension of quality, was mentioned in two of the papers (Montorzi *et al* 2010; Nasser *et al* 2013). This was defined as ensuring that the priorities maximised health for the greatest number, with focus on the poorest. This was not included with the three criteria above as this was viewed more as an externally imposed criterion for determining the quality of the final priorities rather than a mark of the quality of the process.

Viergever *et al* (2010) gave the most detailed description of what a priority setting exercise should involve, giving a detailed checklist of nine points of good practice divided into three phases (Table 3). The purpose of this checklist was to help priority setters "make an informed choice" in choosing a pre-set approach or to help create a high-quality process of their own. It is worthwhile noting that this checklist was tested in global priority setting and that Viergever *et al* (2010) do state that exercises require a degree of pragmatism in considering what works for different situations, stakeholders and with given resources available.

Preparatory work:
- Determine context of exercise and resources available
- Use a comprehensive approach, either pre-set or own methods
- Decide who should be included (inclusiveness)
- Choose what information should be gathered to inform the exercise
- Plan for implementation

Decide priorities:
- Select criteria for setting priorities: three dimensions – public health benefit, feasibility, cost
- Choose methods for deciding priorities

After priorities set:
- Evaluate the priorities
- Write a clear report

Table 3 The Viergever et al (2010) priority setting checklist

Campbell (2010) highlighted the political nature of exercises, which again emphasises the need to consider who to include, and the importance of process and clarity of reasons. In some cases, it may be that the deliberation by stakeholders is more important than the priorities set. This emphasis on mutual listening and learning relies on the ability to incorporate and reflect

different stakeholder voices and thus may require an interpretive approach (Campbell 2010; Lomas *et al* 2003).

Four of the overview publications focused on research priority setting at the global, regional or national level, where governments or institutions (such as the World Health Organisation or the Council on Health Research for Development (COHRED)) set overarching priorities, rather than those in a specific therapeutic area (Campbell 2010; Montorzi *et al* 2010; Oliver *et al* 2004; Viergever *et al* 2010). All except for Oliver *et al* (2004) focus on priority setting for low- and middle-income countries. Thus, some of their recommendations for compound approaches and methods for priority setting may not be as suitable for the scope of the exercise considered in this thesis. In order to consider further what would be suitable for the context of this thesis, examples of actual exercises from other health fields with a similar scope will be considered next.

Authors and Paper	Objective	Method	Key knowledge summary	Indicators of quality
Abma A & Broerse J. (2010)	Develop and examine "a methodology for health research agenda setting processes grounded in the notion of participation as dialogue."	Review and discussion of case studies	The Dialogue Model: six-phase approach grounded in participatory and interactive approaches. Emphasises mutual learning by means of ongoing dialogue and the development of shared action agendas supported by the different stakeholders.	Credibility: participants should recognise the results; enhanced through member check and triangulation. Fairness: enable relevant stakeholders to participate in the process; visibly include their voice.
Campbell, S. (2010)	Examine research priority setting at the national level and at a theme/issue-based level (researchable issues or research questions)	Teaching module	Priority setting is a philosophical, economic and ultimately political act. Deliberating priorities by an inclusive, representative group is more important than setting them.	Adopts the Accountability for Reasonableness framework: "a transparent, fair, legitimate and accountable process."
Cowan K & Oliver S. (2013)	"Step-by-step guidance to establishing and managing Priority Setting Partnerships."	Guidebook	Outlines several generic methods for establishing priorities; defines its PSP methodology.	Transparency of process Balanced inclusion of patient, carer and clinician perspectives Exclusion of non-clinician researchers and organisations with competing interests Audit trail of original uncertainties to final priorities.
Lomas J, Fulop N, Gagner D, & Allen P. (2003)	"… describe how the lessons learned from and the approaches to setting priorities for funding health services are translated into setting priorities for funding health services research"	Review of case studies	Interpretive approaches are more flexible and more able to reflect stakeholder voices. Listening model of 6 steps: identify stakeholders, assemble data, stakeholder consultation to identify issues; validate priority issues; translate issues into themes; validate themes.	"Preventing domination by any particular interest, being transparent, and reflecting the values of all relevant users"
Montorzi G, de Haan S, IJsselmuiden C. (2010)	"Structured as a comprehensive guide that will help the users in designing the most appropriate priority setting process for their countries."	Brief from COHRED experience	Six key practical steps: *"1 Assessing the situation 2 Setting the scene 3 Choosing the best method 4 Planning priority setting 5 Setting priorities 6 Making priorities work"* No 'one best method' for priority setting. Advises weighing complexity of methods	Priority setting is a 'political' process. It must be fair and be seen to be fair. This requires: Capacity and quality assurance; Prioritisation based on evidence, reasons, principles;

			against what is to be achieved and what resources are available. Consider use of more than one method to optimise the usefulness of results.	Transparency by clear documentation and communication; Inclusiveness: all interested parties represented; Promotion of equity, maximise health for the greatest number, with focus on the poorest.
Nasser, M., Ueffing, E., Welch, V. & Tugwell, P. (2013)	To develop a lens to help researchers develop a more equity-oriented approach toward priority and agenda setting in systematic reviews.	Discussion workshop, review of literature on the topic	Developed tool for use in developing a new priority-setting project, prospectively/retrospectively evaluating those processes or comparing different priority-setting methods.	Transparency, inclusiveness, and consider health equity
Nasser, M., Welch, V. *et al* (2013)	Find evidence on the different approaches to research priority setting	Literature review	Gaps in evidence on priority setting methodology: the impact of group dynamics on outcomes; how to evaluate the quality and success of research priority setting methods.	Steps to improving priority setting: stakeholder involvement; group processes need to support the full participation of stakeholders; transparently reporting the criteria of priority setting.
Oliver, S., Clarke-Jones, L. *et al* (2004)	Examine processes and outcomes of national and regional prioritisation exercises.	Systematic review and interviews	Overcome barriers to meaningful consumer involvement with leadership, purposeful outreach, communication, training and support, building working relationships and building on experience.	Framework for evaluation of degree of consumer involvement.
Stewart, R. & Oliver, S. (2008)	Explore the literature on patients' and clinicians' research priorities; reflect on the work of the JLA in relation to this literature.	Literature review	Patients and clinicians can contribute directly or indirectly to priority setting by giving views or by actual collaboration. Contributions from patients and clinicians vary from general topics to specific research questions. Clinicians and patients are more likely to work separately on identifying research topics than collaboratively.	Is engagement direct or indirect, consultative or collaborative? Authors note that the literature has yet to be appraised for the legitimacy and fairness of participation methods or the quality of any evaluations.
Viergever R, Olifson, S, Ghaffar, A, Terry R. (2010)	To develop a checklist outlining options for different approaches.	Literature search; review WHO RPSEs; expert advice	"One gold standard or best practice is therefore not attainable, nor appropriate. "	Checklist of 9 points of good practice: see Table 3.

Table 4 Research priority setting methodology or best practice publications

2.3.3 Methodologies and methods used in research priority setting exercises

Table 30 (Appendix 3, page 230) summarises the 110 exercises not specific to ENT, Hearing and Balance care included in this review. Only 27 out of the 110 publications on RPSE's explicitly stated their methodology. The remainder were pragmatic and outcome-based, focusing on reporting the priorities rather than the underlying methodology. This was true even when they modified or designed their own processes. Of the 83 publications that only stated the process used to set priorities, 22 reported using the Delphi method. None using this method documented an underlying paradigm; rather the rationale given for its use was that it was pragmatic and had established use in healthcare.

The methodological approaches used by the 27 exercises that did outline them are presented in Table 5. This table gives definitions of each and the processes, methods and stakeholder groups associated with them.

Methodology	Description	Processes and methods	Stakeholders
Qualitative/ Exploratory/ Ethnographic (e.g. Keirns *et al* 2009; Corner *et al* 2007 (overlap with participatory approach); Tong *et al* 2008)	"We all use personal experiences, views about ourselves and others, and an in-depth understanding of the circumstances and social context in which we live to produce sophisticated and idiosyncratic frameworks of knowledge about our health and other aspects of our lives… Well-designed and reported qualitative research about priorities across a range of research areas would provide a more methodologically sound route for incorporation of lay knowledge into funding allocation decisions." (Tong *et al* 2008, p 3212)	Stakeholder Identification/engagement. Gathering research ideas/priorities and reasons for prioritising: focus groups, interviews, community forums, observation of groups. Analysis to develop themes: thematic; case-based ethnographic analysis. Note: importance of assessing reasons for prioritising from different perspectives. Normally these criteria are set by funding bodies/researchers. "These considerations are not necessarily inappropriate, simply incomplete." (Tong *et al* 2008 p 3212).	Patients ('unorganised' rather than expert – Corner *et al* 2007) Organised patient/public groups and charities Healthcare professionals Researchers/scientists Note: Usually few types involved; where multiple were consulted, they were not brought together, rather analysis was used to assess and combine priorities determined in separate groups.
Dialogue model (e.g. Abma 2005; Broerse *et al* 2010; Nierse *et al* 2013)	"The underlying notion is that each stakeholder group has its own interests and values, and that the inquirer, instead of preordaining the evaluation by formulating evaluation criteria in advance, should acknowledge this plurality. Methodologically, the acknowledgment of plurality implies that the design gradually emerges in conversation with the stakeholders." (Abma 2005, p 1314) Grounded in participatory, interactive traditions	Identify/engage stakeholders Identify research needs: focus groups, interviews, conversations Prioritisation by separate stakeholder groups: survey, Delphi rounds, focus groups, expert group Integrate stakeholder group priorities to form agenda: dialogue meeting Design programme of work with stakeholders Implementation - call for proposals	Patients/carers Healthcare professionals Researchers Managers Note: Divides stakeholders into two groups - patients and professionals
Participatory /Action (e.g. Cheyne *et al* 2013; Wright *et al* 2006)	"Participatory research is seated in many different philosophical traditions but has at its root the principle of research as a democratic process in	Identify and engage stakeholders, the participatory research group who: • explore focus of study, develop methods;	Patients/carers Healthcare professionals Researchers

	which participants occupy the role of an active citizen within the study." (Wright *et al* 2006, p 7)	• become co-subjects, group methods, such as focus groups most commonly used to identify ideas; • develop new insights by study immersion – focus groups to iterate and refine ideas • further direct study by reflecting on data – analysis to refine lists, determine priorities	Note: Focus is on active involvement and collaboration with patients, often with later integration with other groups
Mixed methodology (e.g. Banfield *et al* 2011; Gagliardi *et al* 2008; Jagsi *et al* 2012; Jensen *et al* 2011; Lowry *et al* 2012)	"Methods that quantify or model the burden of disease or impact of a practice have been poorly adopted because they are resource-intensive, dependent on detailed data, and fail to meet multiple objectives of health systems and their stakeholders. Consensus methods… can be used when data is lacking but… there has been little empirical research to develop these strategies. A mixed-methods approach has therefore been recommended..." (Gagliardi *et al* 2008, p 54)	Identify list of research gaps: Quant: Systematic reviews; Qual: interviews, focus groups, surveys Develop shorter list: Quant: rating survey, literature review for comparison; qual: facilitated discussion, thematic analysis Determine priorities: Quant: ranking survey (may be Delphi round with previous step); qual: facilitated group deliberation	Patients ("consumers") Healthcare professionals Researchers Government Industry Payers Educators Regulators Policymakers (Note: Banfield *et al* 2011 only consulted "consumers")
Priority setting partnership (e.g. Batchelor *et al* 2013; Buckley *et al* 2010)	"Traditionally, the research agenda has been set in a rather uncoordinated manner by the pharmaceutical industry and academia. However, the priorities of these groups may be very different from those of patients and healthcare professionals (HCPs)… The JLA, funded by the National Institute of Health Research (NIHR) and the Medical Research Council, facilitates cooperation between patients and clinicians to identify important treatment uncertainties, thereby influencing the prioritization of future research." (Batchelor *et al* 2013, p 578)	Identify and engage stakeholders: (patient and clinician partnership; set up steering group) Identify treatment uncertainties (survey) Collate uncertainties and refine into questions (by steering committee) Verify uncertainties (literature reviews) Rank into priorities (2 stages: survey followed by Consensus meeting using nominal group process)	Patients Carers Healthcare professionals Targeted exclusion of industry, "pure" researchers

Table 5 Methodologies of RPSEs with definitions, processes and methods, and stakeholders

On examining the overall exercises and Table 5, the following points were considered:

Processes: There are similar stages to the processes adopted by the RPSEs despite using different methodologies (see Figure 3).

Identify and engage stakeholders	•Most methodologies have this as first step •Identification may be iterative; engagement may involve forming steering or research groups
Identify or gather ideas	•May be the first step where systematic reviews are used to identify gaps, then stakeholders identified •Wide variety of methods depending on methodology
Develop list	•Lists of ideas developed through collation + analysis; may be specific questions or broad themes •Some stop here with list of identified themes and do not prioritise further
Prioritise	•Long lists of ideas shortened by comparison with evidence, discussion, rating survey or combination •Priorities set by variety of methods depending on methodology

Figure 3 Process overview from methodologies

Stakeholders: There was wide variation in the reported number of stakeholders involved in the included exercises (see Table 6). Reviewing the lists of stakeholders in the publications showed a disparity in how they were reported. Some authors referred to a list of individual organisations when reporting stakeholder groups (for example, Krishnan *et al* (2013) who stated that they involved "54 stakeholder groups"). Others considered groups as categories of types of stakeholder (e.g. Batchelor *et al* 2013, who involved patients, carers and healthcare professionals).

Range of stakeholders	Number of RPSEs
1	35
2 - 4	27
5 - 10	30
>10	8
Not recorded	10

Table 6 Range of stakeholders reported in the RPSEs (n=110)

Methods: Multiple methods for gathering and determining priorities were used (see Table 7). Most of RPSEs used a combination of methods at different stages of the process. The choice of method was dictated by:

- Choice of methodology, if stated;

- Practical considerations, such as previous use in similar exercises, resource available for the exercise and the occurrence of large stakeholder gatherings such as scientific conferences.

Method	Stage of process used		
	Gather ideas	Develop list	Prioritise
Expert forum/roundtable/working group	✓	✓	✓
Survey	✓	✓	✓
Workshops	✓	✓	✓
Focus groups	✓	✓	
Literature review	✓	✓	
Delphi		✓	✓
Facilitated discussion (small or large group at conference/meeting)		✓	✓
Interviews	✓		
Observation of external groups	✓		
Brainstorm session	✓		
Document analysis	✓		
Scoping meeting	✓		
Nominal Group Process			✓
Dialogue meeting			✓

Table 7 Methods used in research priority setting exercises

Criteria for prioritisation: 28 of the published research priority setting exercises explicitly stated their criteria for prioritisation. There were different approaches to determining these criteria: some were pre-set for the exercise (n=20), whereas other exercises inductively developed internal values or reasons for prioritisation from their stakeholders (n=8). Table 8 summarises the prioritisation criteria inductively derived from stakeholders in those eight exercises; these were often reported through the description of the analysis in the text, as opposed to a reported list of criteria.

Paper	Stated criteria, reasons or values
Criteria stated and derived from stakeholders	
Abma (2005)	Stakeholder discussion provides narrative through text.
Chalkidou *et al* (2009)	(i) Impact on patient health/outcomes, and the intervention effectiveness compared to available alternatives; (ii) Current and projected use of the intervention: variation in practice and diffusion rates; (iii) Safety concerns; (iv) Quantity and quality of the research so far including systematic reviews and research currently planned or in progress; (v) Most appropriate research design and feasibility of research, including costs, randomization issues, and timing, particularly in relation to fast evolving or diffusing technologies; and (vi) Uncertainty surrounding the use of the intervention, particularly in population subgroups (e.g., by age, gender, ethnicity, comorbidities, and so on).
Corner *et al* (2007)	The transcripts provided contextual data regarding the meaning of research topics as well as participants' rationale for identifying them as important.
Foster *et al* (2009)	Reasons embedded in write up - collaborative approach to research; cost burden; need to improve methodology to improve RCTs.
Jagsi *et al* (2012)	Rationale embedded in the explanation of the topic areas: evidence lack; lack leads to underuse hence current unmet need; impact on quality and safety of care; increasing burden of disease hence future unmet need potential; likelihood of improving individual and population healthcare outcomes.
Kierns *et al* (2009)	Acknowledges the importance of different perspectives. Notes priorities based on (1) likely impact on the problem overall; (2) the specific needs of their constituents or communities; (3) their own organizational strengths and skills; and/or (4) larger goals such as health promotion, environmental justice, and community empowerment.
Tong *et al* (2008)	Five reasons participants used to explain their choice for research topics (these were not mutually exclusive): normalization of life, altruism, economic efficiency, personal concerns and clinical outcomes.
Yassi *et al* (2005)	Stakeholder discussion fully analysed to provide narrative through text.

Table 8 RPSEs with criteria for prioritisation determined by the stakeholders

2.3.4 RPSEs specific to ENT, Hearing and Balance Care

Three research priority setting exercises specific to ENT, Hearing and Balance care were identified in the included publications. Two of these were set in the UK (Hall *et al* 2013, JLA Balance PSP 2011). The third had been done in the Netherlands (van Benthem *et al* 2013).

Table 9 outlines the three exercises giving their scope, stakeholders, methodology, methods and outputs. The priorities for these exercises may be seen in Tables 31 and 32 in Appendix 4, page 236.

None of these exercises addressed the full scope of the research question in this thesis. The two exercises from the UK (Hall *et al* 2013; JLA PSP 2011) used the established Priority Setting Partnership Methodology. The scope of these exercises was however limited to a single topic, and the range of stakeholders included in the exercise was limited in order to focus on patient and clinician priorities.

The third exercise (van Benthem *et al* 2013) covered the broader specialties as opposed to focusing on a defined clinical topic. It developed its own process, which was clearly reported and used established methods, although underlying methodology was not obviously discussed. This exercise was confined to clinicians and patient representatives.

RSPE and Year	Country	Scope	Stakeholders	Methodology + methods	Outputs
James Lind Alliance Balance Priority Setting Partnership (2011)	UK	Topic-specific: balance	Patients Clinicians	Priority-setting partnership methodology: Identify/engage stakeholders; Survey to collect uncertainties; Uncertainties refined and collated; Uncertainties verified by comparison with the literature; Interim prioritisation by ranking survey; Final prioritisation in Consensus workshop using Nominal Group Process.	Top ten research questions No prioritisation criteria given
James Lind Alliance Tinnitus Priority Setting Partnership, Hall *et al* (2013)	UK	Topic-specific: tinnitus	Patients Clinicians	Priority-setting partnership methodology as above.	Top ten research questions No prioritisation criteria given
De KNO-wetenschapsagenda, van Bentham *et al* (2013)	The Netherlands	ENT (whole specialty)	ENT physicians Patient organisations	Developed own methodology: Identify knowledge gaps from database search, guideline analysis and survey of participants; Prioritisation by Consensus Conference participants through scoring using pre-defined criteria.	53 knowledge gaps in three categories (primary research questions, guidelines, other) divided into ENT sub-specialty areas. Criteria for prioritisation given: relevance (severity, prevalence, costs); urgency; researchability / feasibility; impact on the field / society

Table 9 ENT, Hearing and Balance specific RPSE

2.4 Discussion

2.4.1 Choosing methodology and process

This review shows that quality of the process used to determine priorities is important in developing a research priority setting exercise. It also provides us with a tripartite lens through which to determine that quality – is the process transparent, inclusive and credible? – as well as providing the Viergever *et al* (2010) checklist. That lens will now be used to review the findings to determine how to design a RPSE for this study.

Methodology: Several established and emergent methodologies were identified from the exercises. Were any of these suitable to answer the research question of the thesis? As outlined in Chapter One, the context here is to develop an exercise that:

- Covers all of ENT, Hearing and Balance care;
- Incorporates the views and voices of multiple stakeholders.

Given that two previous UK exercises used Priority Setting Partnership methodology this would seem to be a good choice as it is an established process. Viergever *et al* (2010) recommended using an existing approach if it was appropriate for the context of an exercise. However, targeted exclusion of some stakeholders is fundamental to this methodology, making it unsuitable

to answer a research question designed for wider inclusion. Instead, the other methodologies outlined in Table 5 were considered.

The first methodology in that table is the qualitative approach, described as a methodologically sound way to incorporate different voices (Tong *et al* 2008). From Table 5 it can be seen that this approach was most often used with a limited spread of stakeholders, and where multiple ones were consulted, this was done separately. Although this allowed stakeholders to have detailed deliberations within their own categories, for example separate discussions for patients and researchers, it did not allow for cross-category deliberation. There is no restriction to combining these different types of groups, but how this was done would need to be carefully considered so that all stakeholders were truly included.

A similar issue arose with the Dialogue Model (Tables 4 and 5) and participatory approaches (Table 5), in that they were seen to be used with a limited spread of stakeholders. The only methodology found in this review that was used with a spread of stakeholders was mixed methodology. Gagliardi *et al* (2008) (Table 5 and Table 30, Appendix 3, p 230) used mixed methodology and it is interesting to examine their exercise in greater detail. The authors combined technical quantitative data (ranking and rating to identify and prioritise research gaps) and interpretive qualitative strategies (workshop deliberation about prioritised gaps and development of research questions) to identify and prioritise colorectal cancer health services research questions. This was done to allowed them to capitalise on both the qualitative and

quantitative strategies. However, this exercise demonstrated a weakness with using this combined approach. In the interactive workshop discussion to deliberate the prioritised gaps participants felt that there was a need to "generate a common understanding, particularly with respect to the clinical issues and nature of research being considered" (Gagliardi *et al* p 59). Discussing pre-determined topics of research identified through quantitative methods did not create this shared meaning. This exercise achieved its immediate objectives, insofar as gaps in knowledge were identified and consensus reached on research priorities. However, participants noted a lack of common understanding of the nature of the research being proposed, and on follow up no projects based on the results had been planned by participants. What Gagliardi *et al* (2008) demonstrated was that mixed approaches may not address an aim of reaching shared understanding and meaning with different stakeholders, thereby capturing their views and voices. To do this a more exclusive qualitative approach is needed.

Process: With the Dialogue Model and Priority Setting Methodology (Table 5), the process was pre-set into distinct steps. The other methodologies were not as prescriptive, but as can be seen in Figure 3 above, an overarching process can be derived from the different approaches. This is used to provide a framework suited to the context of the exercise here. Determining who to include and how to include them is more complex given the range of stakeholders and variety of methods used in exercises (see Tables 6 and 7). This is considered in the next section.

2.4.2 Stakeholders and methods

To ensure stakeholder inclusion in the design of the exercise for this study, the following points were considered:

- what was meant by "stakeholders";

- who they should be;

- and what methods should be used to include them.

Stakeholders: are defined in the literature as: "any group, individual or institution with a vested interest in the prioritisation process" (Campbell 2010, p 33). Nasser, Welch *et al* (2013) adopted Tugwell *et al*'s (2006) categories of the "6 Ps": public, patient, practitioner, policy maker, press, and private sector, with an added seventh stakeholder being the researchers themselves. This categorisation is useful given the lack of common terminology seen in the literature, with some exercises considering stakeholders by category and others considering stakeholders as individuals or organisations.

This is an important distinction to make. Exercises that claim that they involve multiple stakeholders representing a range of views may be representing the views of a single category. As this exercise aims to incorporate the views of multiple stakeholders (as discussed in Chapter 1), stakeholder category mapping, clearly defining the categories to be engaged, is used as the first step in the exercise to ensure that there is actual broad inclusion.

Who should be included: Historically patients and carers have not been consulted in research priority setting (Cowan and Oliver 2013). Involving them is now seen to have a political and practical mandate, as they help design

"better," more relevant research on which to base health services (Caron-Flinterman *et al* 2005). There have been objections to including these non-traditional stakeholders. These concerns are that individuals lack knowledge and understanding of scientific language and are unable to objectively set priorities uninfluenced by personal experience and the media (Caron-Flinterman *et al* 2005). As a result, the choice of stakeholders and how they were recruited has developed in a number of different ways, as outlined in the section considering methods below.

Some exercises tried to redress the historical power imbalance in priority setting by targeted exclusion of industry and non-clinical academics (Cowan and Oliver 2013; Table 4). This was the approach adopted by the previous ENT, Hearing and Balance exercises that used priority-setting methodology (Hall *et al* 2013; JLA PSP 2011; Table 9) or adopted a similar model (van Benthem *et al* 2013; Table 9). The danger here was that inclusiveness was seen through the narrowed lens of consumer involvement only. Broad stakeholder involvement has been seen as generally beneficial as it prevents research options from being overlooked, fosters ownership, improves implementation of priorities and may prevent unnecessary duplication of exercises (Viergever *et al* 2010). The other danger was that focusing exclusively on patients as "users of research" may make that definition too narrow. Lomas *et al* (2003) noted that:

"In the case of research, however, the users include not only patients and providers but also managers and policymakers striving to make the best

possible decisions in an evidence-based health service" (Lomas *et al* (2003) page 364, citing Walshe and Rundall (2001)).

Whether the view of "users" alone is adequate to develop a national research agenda may also be debated. Some of the exercises, such as Saldanha *et al* (2013; Table 30, Appendix 3, page 230), note that there is a danger that the exclusion of traditional experts, such as experienced researchers, may result in a lack of clarity and feasibility in the output. The "expert" may be needed for context, providing understanding as to why and how far the research gap that has been highlighted exists. To develop a balanced agenda that reflects national priorities, is feasible and mobilises existing research capability to adopt the agenda, the views of stakeholder groups beyond patients, carers and healthcare professionals should be given consideration.

Methods to include stakeholders: Alongside methodological considerations, how methods may be used or combined to promote stakeholder inclusion is important for fairness of process. Patient and carer stakeholders are at greater risk of poor inclusion (Stewart and Oliver 2008; Table 4). This may be due to difficulty in recruitment or challenges in ensuring their views are heard due their lack of experience in research compared to more traditional stakeholders.

Different methods were suggested to overcome this (see Tables 5 and 7). One way of ensuring inclusion is through the recruitment of "organised" patient representatives. Here, patients and the public are represented by

experienced, empowered civil organisations (Campbell 2010) that already have expertise and experience in research. Using "organised" interest groups (Corner *et al* 2007) redresses the power imbalance between the "scientific" and "lay" community and allows for meaningful dialogue and deliberation. The concern with the organised representative model is that, although it addresses the problem of power imbalance between groups, it does so at the risk of ignoring the "silent voices" (Abma 2005, p 1324). This means that these organised groups may lack diversity and not be truly representative or inclusive, especially with regard to marginalised groups. Thus, standard methods of data gathering, such as surveys, may be useful to reach large numbers, but Abma (2005) noted these methods of self-selection may fail to gather the views of a range of patient stakeholders. Some of the exercises were seen to address this by using more than one method of information gathering, such as narrative workshops and conversational interviews (Abma 2005) or focus groups and questionnaires (Caron-Flinterman *et al* 2005). Other exercises deliberately sampled from marginal groups, for example Corner *et al* (2007) where they used purposive sampling for focus groups.

More subtle means of exclusion may also exist. As Nasser, Welch *et al* (2013; Table 4) noted, simply being present in the meeting does not mean that exclusion does not happen, especially where there is the use of technical jargon. In mixed stakeholder groups the very language used to discuss research priorities may give rise to exclusion, as more deference is generally given to the language of traditional experts such as researchers (Abma 2005; Table 5). This can subtly devalue the experiential expertise of patient or lay

stakeholders and exclude them from true participation in the discussion. Participants in research priority setting have been noted to value the opportunity to engage in discussion in group settings, so it was important to avoid further marginalisation if using these methods (Tong *et al* 2008; Tables 5 and 29 Appendix 3, page 230).

Subtle exclusion is about more than ensuring that technical language is not overused in group discussions. It underlines the importance of creating a shared language around research and why it is important, a point emphasised by Abma and Broerse (2010). To do this, creating a mutual understanding of what is meant by research by all the stakeholders needs to be considered, especially when considering the views of 'unorganised' patients. Inductively developing a common framework from the views of stakeholders may facilitate this understanding.

2.4.3 Methods and different steps of the process

Table 7 highlights the many methods employed at all stages in the exercises, chosen either for methodological or pragmatic reasons. Availability of resources for RSPEs is a key component of determining methods (Montorzi *et al* 2010) as they will have different resource requirements. A survey may be an imperfect but useful method as it uses fewer resources than large group meetings or conferences. Many of the exercises that used resource-intensive large group formats also had small group or breakaway sessions to benefit from focused discussions as well as large group consensus in one sitting.

Credibility, identified as an indication of quality of process in the overview papers, also plays a role in choice of methods at each stage in designing the exercise for this thesis. The use of more than one method and multiple stages optimises the usefulness of results and allow for triangulation, (Montorzi *et al* 2010; Abma and Broerse 2010). Methods that allow for deliberation and assessment of results by stakeholders throughout the process were used to enhance credibility in the design of this study, as an alternative to sending out lists of determined priorities only at the end of the exercise. When these discussions are transparently reported, they also provide an audit trail for the decisions that are made.

2.4.4 Determining and reporting criteria for prioritisation

Transparency was identified as a crucial indicator of quality in this review. One of the underreported areas noted in section 2.3.3, page 52, was in capturing the criteria, reasons or values that informed the prioritisation decisions.

Nasser, Welch *et al* (2013) stated that values need to be considered in priority setting, especially if true and diverse involvement is to be ensured. Campbell (2010) concurred and viewed agreement for criteria for specific exercises to be a critical step, an opinion shared by Viergever *et al* (2010) and Montorzi *et al* (2010). How these criteria are determined is less straightforward. Viergever *et al* (2010) stated that these should be determined by consensus at the

beginning of the exercise, and outlined that they usually fell into one of three domains:

- Public health benefit (should we do it?);

- Feasibility (can we do it?);

- Cost.

The exercises described on page 52 and those listed in Table 8 did not necessarily follow the approach advocated by Viergever *et al* (2010). As in much else in priority setting, this would appear to be less about "right and wrong" and more about what will answer the aims of the exercise. Some exercises took a pragmatic approach, noting that pre-set criteria allowed them to align the priority setting values with those of funding organisations or the bodies sponsoring the exercise (for example, Krumholz *et al* (2005)). Whether these reflect wider values in multi-stakeholder exercises has been less explored.

This was discussed in Tong *et al* (2008) where the lack of examination of different stakeholder reasons for prioritisation was noted. In that exercise the focus was on patient-derived reasons which were determined from focus group analysis. The criteria for prioritisation highlighted in that approach were: normalisation of life, altruism, economic efficiency, personal concerns and clinical outcomes. The reasons do not all fit neatly into the Viergever domains, and highlights that with non-traditional stakeholders a different approach may be needed. Furthermore, if the discussion is considered as important as the

final output, then an inductive approach to developing these values from participants through the exercise will more clearly reflect the views and voices of a wider range of stakeholders. Inductively developing values in an exercise does not mean that there will be complete agreement on them. Where there is lack of consensus this too should be captured and reported in order to be transparent.

2.5 Strengths and limitations of the review

This review used a robust method suitable for assessing diverse and emergent literature, which was a strength of the approach taken. This review did have some limitations. Publications were not assessed for quality or risk of bias. The focus of the review was to provide breadth rather than depth of information, therefore some areas, such as a greater understanding of the methods used, could be further developed. Only one researcher reviewed all the papers and developed the themes described; two independent reviewers would have been a more robust approach, but this resource was not available.

2.6 Conclusions

The findings and discussion above will be used to design a robust exercise that balances pragmatism and quality in the complex setting of a multi-stakeholder approach. Scoping the literature suggested that in order to

incorporate the views and voices of multiple stakeholders in a research priority-setting exercise the methods need to start with mapping the stakeholders. Determination of who exactly the stakeholders are through developing stakeholder categories also leads to the choice of the methods used. These need to promote stakeholder inclusion so that expertise and experience from both traditional and non-traditional experts are incorporated. Given these different perspectives, the deliberation around the priorities is as important as the final list in order to reach this shared understanding.

To make sure that their voices are heard without exclusion, qualitative methodology in a multi-step process using a variety of methods is the most robust approach to develop shared meaning between stakeholders.

There is also a need to transparently report not just the process, but also the underlying reasons and discussions about why the priorities are chosen. Without this there may be a danger that stakeholders are assumed to share common values and viewpoints. This is important to explore as the assumption that these spoke with a common voice could result in the subtle exclusions that were not appreciated in some of the documented exercises.

This chapter has used the findings from the scoping review to outline how to design a research priority setting exercise based on the views and voices of its stakeholders. The next chapter details the methodology, process and methods developed for this study.

Chapter 3: Methodology

3.1 Introduction

This chapter details the methodology, process and methods used to answer the research question "What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?" In Chapter 2, the scoping review showed that there had been research priority setting exercises (RPSEs) in topic specific areas of UK ENT, Hearing and Balance care. These were limited exercises both in the range of stakeholders consulted and by the fact that they only addressed a specific clinical topic. As stated in that chapter, the process used by these exercises was not suitable here, therefore another approach was adapted. This balanced the principles outlined by Viergever *et al* (2010) that suggested that using established methodology may improve the quality of the exercise, but pragmatically adapting it to suit the context of this exercise.

3.2 Process and Methodology

This exercise was designed as a multi-stage process (see Figure 4). This was based on the findings from the previous chapter. It provided a staged approach with continual engagement of a broad range of stakeholders. In Chapters 1 and 2 it was noted that broad stakeholder engagement would be beneficial to the exercise. In choosing this broad range, the challenge was to

ensure that the voices of the stakeholders were neither excluded nor became too dominant in the development of priorities. To overcome this obstacle, consideration was given as to how to develop shared meaning.

In this exercise qualitative methodology was used to address this challenge. The previous chapter found this was more effective in developing shared meaning from the views of a range of stakeholders than mixed methodologies. This also informed how data were analysed. An inductive approach was used to develop priorities from stakeholders, rather than using a deductive approach starting with a pre-set hypothesis and testing it.

The next section outlines the methods used in each stage of the process.

Figure 4 Overview of the GENERATE process and stages

Steering Committee

Survey

Focus groups

Expert Forum

Consensus Conference

UK ENT Hearing and Balance research agenda
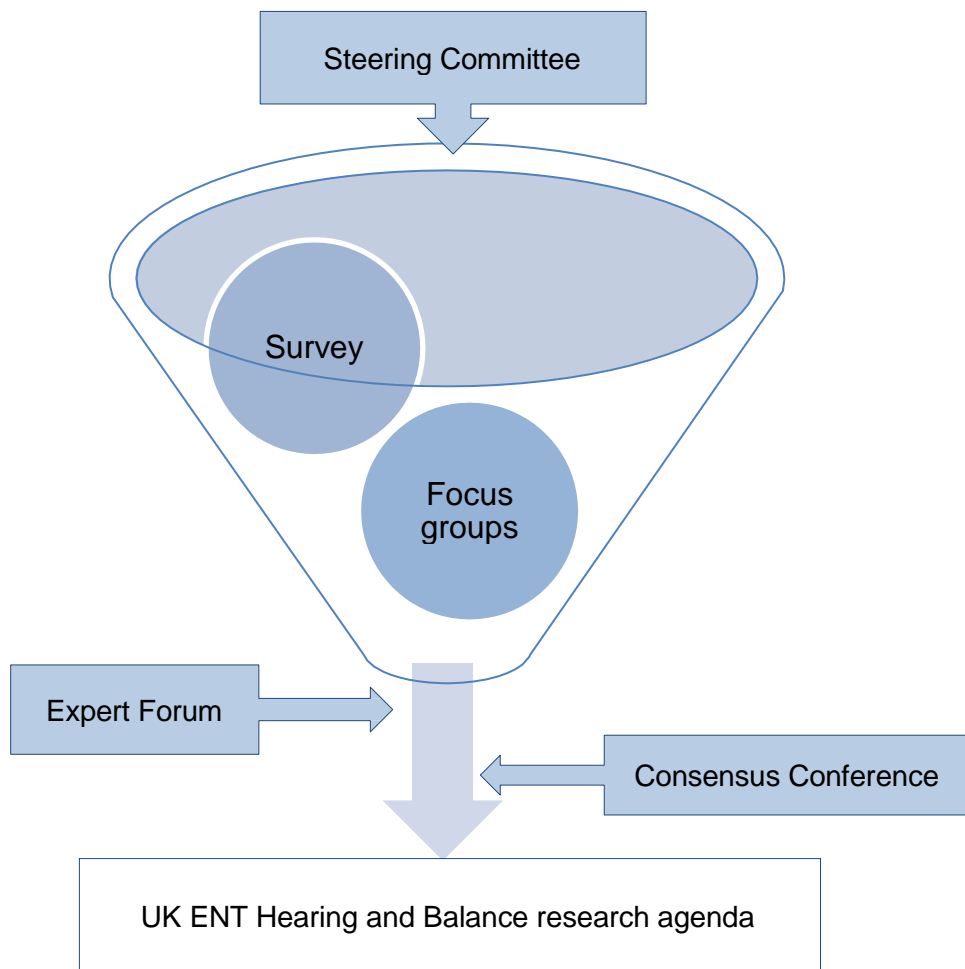
Process stages

1. Identify stakeholders

2. Gather research ideas

3. Analyse ideas

4. Develop final list of priorities

3.3 Methods

3.3.1 Stakeholder Identification

From the literature review, stakeholders were defined as: "any group, individual or institution with a vested interest in the prioritisation process" (Campbell 2010, p 33). In order to define the sample population for this study and answer the question "who are the stakeholders for UK ENT, Hearing and Balance care", stakeholder categories were developed with input from the relevant literature and from expert opinion.

This exercise was designed to be comprehensive, so that multiple stakeholders' views were incorporated to form the final priorities. To ensure that potential stakeholders were not overlooked, a hierarchical approach with the initial identification of overarching stakeholder categories (Tugwell *et al* (2006)) was used (see Ch 2, page 59). These categories were then populated by relevant groups and individuals. Both the categories and lists of individual stakeholders were refined and expanded iteratively.

Initial stakeholder categories were based on the findings of the literature review (Lomas *et al* (2003); Cowan and Oliver (2013); and Saldanha *et al* (2013)). In addition, there was discussion with organisers of other RPSEs to understand what stakeholder categories they had worked with, why they were chosen and the challenges in working with them.

Following this initial identification, a Steering Committee was formed with members recruited from the stakeholder categories. These categories were refined further in discussion with the Steering Committee to produce a stakeholder map for the RPSE.

The Steering Committee was a group of 20 stakeholders, with a neutral Chair from outside the field of ENT, Hearing and Balance care. The role of this group was to agree the final process for the RPSE, provide advice and networks to engage stakeholders, and comment on the analysis and output.

Having determined who the stakeholders were for the exercise, methods then needed to be developed to determine their priorities and why they chose them.

3.3.2 Methods of collecting research ideas

Two methods of collecting research ideas were used: a survey and focus groups. The use of more than one method was noted to optimise the usefulness of results and allow for triangulation (Ch 2 page 64). A survey was used as it was a pragmatic method of gathering ideas from a broad range of diverse groups (Viergever *et al* 2010). Focus groups were chosen as the second method to collect ideas as they:

- allowed for greater understanding of the perspectives of different populations, especially those underrepresented in the survey or difficult to access (Abma 2005);

- provided participants in research priority setting the opportunity to engage in groups, which they are reported to value (Tong *et al* 2008).

Previously identified research ideas, such as recommendations from existing professional guidelines, were not directly used as a source for this study. This was to avoid the potential for subtle exclusion of stakeholder categories, particularly patient and carer voices, who had not been able to input into their development. Using these would have put too much weight of response into ideas that came from categories that traditionally set priorities. Number of responses did not form part of the data analysis (see section 3.3.3 below). Nevertheless, having a large volume of technical ideas as part of the data risked skewing the inductive development of ideas from all stakeholder voices. If these truly represented priorities from the clinical community, they expectation was that they would be suggested by the stakeholders through the survey and focus groups.

3.3.2.1 Survey

Sampling: There was no specified sample size for the survey; rather a large number of responses was sought. Participants were selected through convenience sampling, a pragmatic technique that allowed for widespread distribution and engagement (Broerse *et al* 2010). Responses were sought from:

- Categories from the stakeholder map;

- All four nations of the UK;

- A range of age groups;

- All genders;

- A range of ethnic groups.

Survey Instrument: The survey instrument was based on validated open-response surveys used in other priority setting exercises (Sight Loss and Vision Priority Setting Partnership 2014, Howell *et al* 2012) and adapted for this exercise. Different formats for the questions were considered, including the PICO format (population, intervention, comparison and outcome) used to develop research questions. It was decided that using an open question format allowed respondents from a range of backgrounds to articulate research ideas in their own words.

The survey instrument was piloted with representatives from the stakeholder categories. Following feedback minor adjustments were made to the introductory text in the survey, placing greater emphasis on responses being based on personal experience, clarifying the scope and ensuring the language used was suitable for a range of stakeholders.

The final survey consisted of an introduction explaining the purpose and scope of the survey, and two free text questions:

- What do you think are the most important topics, questions or issues that should be answered by research for ENT, Hearing and Balance?

- Why do you think these are important?

The survey was anonymous, but respondents were asked to voluntarily provide demographic data in order to assess the spread of respondents. A copy of the survey instrument may be seen in Appendix 5, page 239.

Survey dissemination: Table 10 shows the routes and methods of dissemination of the survey. Appendix 6, page 244 gives further details of organisations contacted, which of these distributed the survey, and meetings and events where it was disseminated.

Route of dissemination	Number contacted	Number disseminating survey	Method of dissemination
Stakeholder organisations	383	59	Publicity articles and electronic link distributed through membership email lists, newsletters, websites and social media
Hospital clinical leads	63	4	Paper copies in outpatient clinics
Conferences and meetings	6	6	Presentations with electronic link and paper copies to delegates
Patient and public events	2	2	Paper copies to attendees

Table 10 Routes and methods of survey dissemination

3.3.3.2 Focus groups

Sampling: Purposive sampling was used. Characteristics considered in the sampling for the focus groups are shown in Table 11. The aim was for each

focus group to consist of six to ten members. Potential participants who were

unable to give consent due to a lack of capacity, as defined by the Mental

Capacity Act 2005, were excluded.

Characteristic	Considerations
Stakeholder	Sample across stakeholder categories, or from underrepresented groups within a category.
Age	Sample a range of ages. Children below the age of consent excluded.
Gender	A balance of male and female participants.
Geographical location	Sample across the four nations of the United Kingdom (subject to resource constraints).
Ethnic origin	A range of ethnic origins to reflect diversity of UK population.

Table 11 Characteristics for focus group sampling

Recruitment: Recruitment was through a variety of sources:

- professional organisations and networks;

- direct contact with individuals on the University College London (UCL) evidENT ENT Patient and Public Involvement (PPI) database;

- advertising, specifically in the People in Research website (a resource open to the public and funded by the NIHR to support active public involvement in NHS, public health and social care research).

Recruitment was not through established patient charities as the survey was

widely publicised through these routes and the focus group allowed the

exploration of the views of "unorganised" individual patients (Corner *et al*

2007). Recruitment of patients was supported by a professional Patient and

Public Involvement (PPI) Coordinator to ensure that national standards for working with patients and the public in research were met.

Method: Topic guides were developed focused on the following areas:

- To generate ideas for priority areas of research in ENT, Hearing and Balance care.

- To explore in greater depth the reasons why participants believed that these were important areas of research and why they should be prioritised over other areas of research.

- To explore what research themes participants felt had emerged from the discussions and use this data to refine data analysis from the survey.

The topic guides may be seen in Appendix 7, page 247.

The focus groups were not used to try to reach consensus over the issues and priorities discussed, but rather to capture diverse and divergent opinions. Information about the objectives of the focus group and the overall study were provided to all participants (see Appendix 8, page 248). Confidentiality and anonymity were assured, and each participant was asked to sign a consent form including permission for audio recording and for results to be used in the research and development of the agenda. They were asked to provide basic demographic data consistent with the data collected in the survey. Each group was moderated by two trained facilitators. The discussions were recorded and independently transcribed for analysis.

3.3.3 Collating/analysing ideas

Survey responses and focus group transcripts were analysed using the Framework Method. Given the broad range of stakeholders consulted and the different methods used, it was anticipated that the analysis would involve a large volume of information expressed in very different ways. The papers in the literature review were not detailed as to the exact methods of analysis used in exercises adopting a qualitative approach. Most stated that they used some form of qualitative or content analysis (Dzidowska *et al* 2010; Sawin *et al* 2012). These described coding practices and the use of independent coders and discussion to resolve disagreements (Tong *et al* 2008; Nierse *et al* 2013). As the papers did not give detailed descriptions of the stages of analysis, an analytic method was chosen to provide this structure.

The use of the Framework Method has not been previously described in the literature relating to research priority setting. It was chosen as it was:

- a systematic approach to qualitative data analysis;

- flexible, not aligned to any particular epistemological approach, that could be used inductively or deductively (Gale *et al* 2013);

- developed for use in policy research (Ritchie and Lewis 2003) and widely used in health research (Gale *et al* 2013);

- a method that organised and analysed large amounts of information systematically and visibly, whilst retaining links to the original data (Ritchie *et al* 2003);

- a dynamic method, allowing for iterative change through the analysis process (Srivastava and Thomson 2008).

The five key stages of analysis for this method (Richie and Spencer 1994) are shown in Figure 5 below and were used in this exercise.

The data were analysed independently by members of the Academic Team to minimise bias. This team consisted of five members from different backgrounds, both clinical and non-clinical. Two members of the team ("Researcher 1," the principal researcher and author of this thesis, and "Researcher 2," a non-clinical post-doctoral researcher) analysed the full dataset. The other three (professors supervising this thesis) analysed different samples of ten percent of the survey responses and two focus group transcripts.

Familiarisation
with the data

Develop
thematic
framework

Indexing: apply
framework to
data

Charting: data charts
with themes as
headings

Mapping
charts to
develop
concepts

Figure 5 The five stages of Framework Analysis (Richie and Spencer 1994)

Analyses were compared, discussed and refined through meetings with the
Academic Team. The frameworks were iteratively developed, following the
principle of "define and refine" (Braun and Clarke 2006). After the initial
development of themes, Researchers 1 and 2 independently indexed the data
into the proposed themes and then created modified case charts for each
theme. These combined similar responses instead of charting each individual
response due to the volume of data.

Iterations of the frameworks were shared with the Steering Committee for their comments, which were used to further refine them. As well as developing the analysis, this was a way of ensuring credibility and transparency by exposing the analysis to peer review and checking it with members of the population with whom the research was being conducted.

The outputs of this stage of the analysis were the framework developed from the responses and a long list of research ideas. The development of the framework underwent multiple iterations as it involved both developing the structure of the framework and the long list of research ideas under the framework headings. The long list of research ideas was then prioritised through the methods described in the next section. Further development of the framework and ideas continued based on feed back through the subsequent prioritisation process. An overview of the process of developing the research ideas framework may be seen in Figure 6.

Figure 6 Framework development process

Key:

R1 – 5: Researcher 1 – 5

SC: Steering Committee

→ Multiple iterations

--→ Single iteration

Expert Forum

Input: survey + focus group data

82

R1

R2

Independent analysis + Team discussion

Themes Topics

R5

R1

R2

R4

R3

Independent analysis + Team discussion

Themes Topics

SC input

Output: Framework
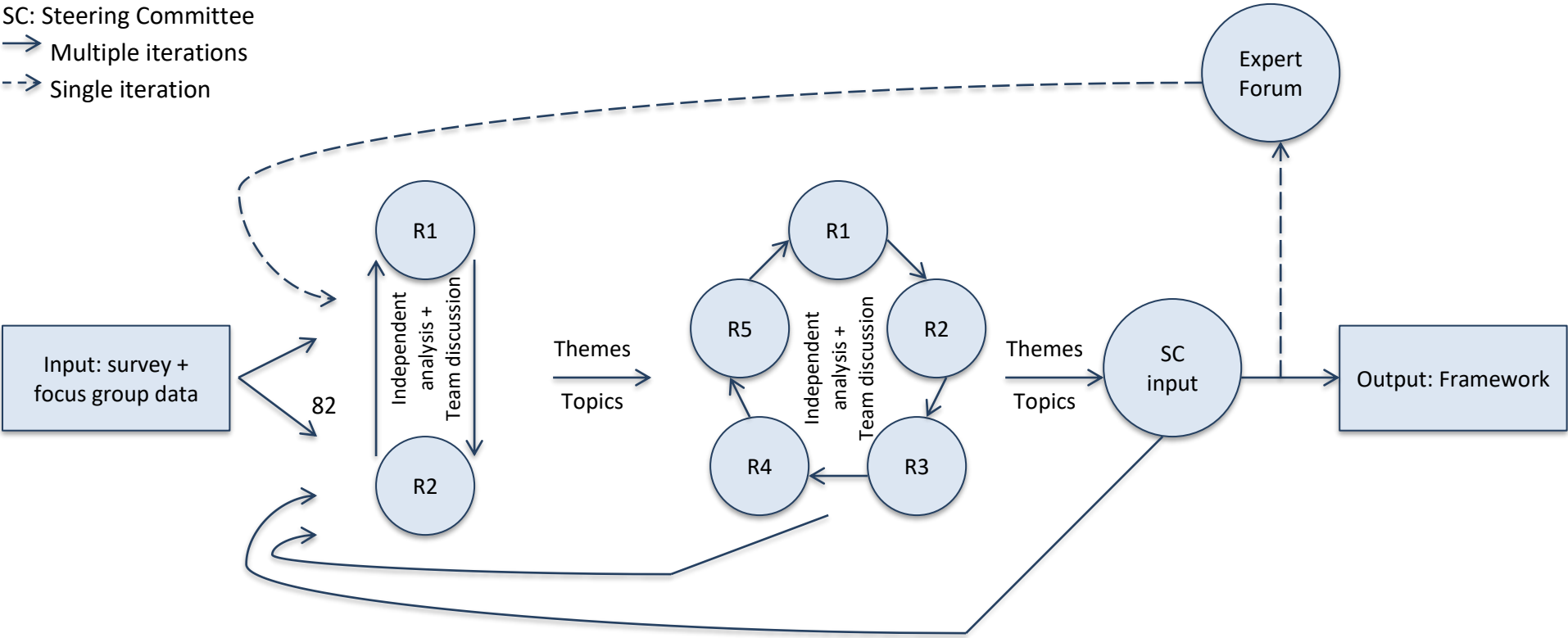
3.3.4 Developing the list of priorities

Developing the list of priorities was a two-stage process based on techniques from the literature review. The first step involved assessing which of the long list of ideas had already been answered or was in the process of being addressed by existing or ongoing research. This led to the development of a shorter list. The second step determined which of the ideas from the shorter list would become the research priorities.

In Chapter 2 it was seen that developing shorter lists could be done by literature reviews, rating surveys or discussion. In this exercise the third option was chosen, and discussion in an expert forum was used. There were several reasons for this choice:

- The use of experts allowed for nuanced discussion around whether the current evidence sufficiently addressed the idea. It was recognised that a literature review would determine if the idea had been researched but did not give insight into whether this evidence was sufficient or was adopted in clinical practice, a concern that was considered in Chapter 1, page 20.
- Given the broad range of stakeholders that were consulted a wide range of responses involving different research fields was expected. Systematic literature reviews for a large number of ideas was not feasible with the resources or timeframe for this exercise. Thus the use of an expert forum was a pragmatic and efficient choice.

- Experts from different disciplines would also be able to give insights as to whether these questions were currently the subject of ongoing research in academic specialties outside of traditional ENT, hearing and balance care.

- Asking experts to take part in a forum meant that their opinions could be challenged and discussed. Although this did not eliminate the risk of individual bias of the experts it provided a way of mitigating it.

A consensus conference was used for the second step. This allowed for selection of the final priorities through a combination of both large group and small group focused discussions with a broad spread of stakeholders.

3.3.4.1 Expert Forum

Sampling: Participants in the Expert Forum were purposively sampled based on academic expertise in the themes and topics that formed the framework for the research ideas, and on their experience of assessing, funding, designing or running high quality studies. As patients and patient representatives were considered "experts" in this exercise, they were included as participants (Howell *et al* 2012). The patient experts were required to have experience in Patient and Public Involvement (PPI) panels or reviewing research proposals. The aim was for the group size to be approximately 20 participants, with two patient experts, so that both small and large group discussions were feasible. Appropriate expertise was considered more important than geographical or demographic diversity.

Recruitment: Clinical and non-clinical academics were recruited based on the recommendation of the Steering Committee and Academic Team after reviewing the draft framework. Patient experts were recruited through established PPI panels.

Method: A briefing document was sent to participants prior to the forum that included the long list of research ideas their consideration (See Table 34, Appendix 9, page 254 for the long list of ideas). Three questions were posed to the participants in this document and in the Forum:

- In their opinion, and based on the current state of evidence, which ideas developed from the data needed more research?
- What type of research would be of highest value and doable in those areas?
- What values and criteria did they use to decide what topics were important to research?

Each participant was asked to sign a consent form including permission for audio recording and for results to be used in the research and development of the agenda. They also completed Declaration of Interest forms.

An independent professional facilitator conducted the Expert Forum. There was a combination of small and large group discussions, with the small group work being fed back and discussed in a plenary session. Small and large group discussions were audio recorded and transcribed. Notes from the small

group discussions were captured by participants on A3 paper copies of the framework and the long list of ideas. Notes from the full group discussion were also made by Researcher 1.

Analysis: Notes and transcripts from the Expert Forum were read and compared with the long list of research ideas. The ideas that the experts felt had been adequately addressed or were being investigated by existing research were removed. Ideas were also combined based on their recommendations. The notes and transcripts were also read to assess suggestions about how the framework could be improved. The Academic Team discussed these suggestions in the context of the data from the survey and focus groups, and further refined the framework. The reasons that the experts gave for their recommendations were recorded throughout the forum. A formal report was drafted following the event and circulated to the participants for their comments to ensure views had been accurately captured (see Appendix 10, page 288).

3.3.4.2 Consensus Conference

Sampling: Participants were purposively sampled to reflect both the stakeholder map, and to include patients and professionals with experience of the themes and topics that formed the framework of research ideas. The proposed sample size was 50 – 60 participants in order to be able to accommodate discussion in a plenary session as well as small group work, with each small group to have six to ten members. Participants were sampled

from across the UK and from a range of ages and ethnicities. At least two patient participants were required for each small group. This was to ensure adequate patient representation in these groups and to prevent domination of the discussion by the other stakeholders.

Recruitment: Recruitment was through professional organisations and networks, established PPI groups and the UCL evidENT PPI database. The latter consisted of patient who were recruited through clinics and public awareness of research events and had given their permission to be contacted for research. An experienced PPI coordinator was responsible for the recruiting patient and public participants, provided training if needed, and acting as their primary point of contact from the point of recruitment onwards.

Method: A briefing document was sent to participants with information about the aims, objectives and methods of GENERATE and the analysis of responses (see Appendix 11, page 315). It included the short list of research ideas agreed after the Expert Forum for prioritisation by the participants, and a discussion of the reasons why these areas had been selected. The tasks for the participants in the conference were stated in this document. These were:

- To discuss what they thought were the most important questions to be included in the agenda.
- Why they thought those were important.
- To come to an agreement as a group at the end of the Conference as to what to include in the final agenda.

Following an opening plenary session, participants took part in small group discussions moderated by trained independent facilitators. A briefing guide was developed for these discussions (see Appendix 12, page 350). Each small group was asked to choose three to four top priorities from their list following individual presentations of ideas and group discussion. Finally, there was a concluding plenary session where the small group work was presented to the whole group for peer review, debate and final consensus through discussion.

All discussions were audio recorded, following participant consent, and independently transcribed. The small groups made written notes on posters provided for display and review by the other participants, and the small group facilitators made brief field notes based on their sessions.

Analysis: Transcripts, participant notes on posters and field notes were reviewed to determine the final priorities in the framework and the reasons for prioritising them. Areas of disagreement and how they were resolved by discussion in the group were also described in the final analysis.

To assess for bias in excluding ideas in these stages both the ideas presented to the Consensus Conference and the final priorities were mapped against the framework. The purpose of this was to see if they were spread across the themes, which had been directly developed from the detailed, rigorous analysis of the data. The rationale for this was that the framework

had been rigorously developed from the whole of the data; if themes were neglected it would indicate that ideas weighty enough in the data to form these themes had been excluded by the smaller group of stakeholders prioritising them. The final priorities were also mapped back to the original data from the survey and focus groups. This was to determine if the views and ideas of a spread of stakeholders were represented, or if ideas from one stakeholder category had taken precedence through the process.

3.4 Summary of quality considerations for the process developed

As was outlined in Chapter 2, page 46, with there being no gold standard process for research priority setting, assuring the quality of the process is critical. The literature identified three aspects of quality that needed to be satisfied: transparency, fairness and credibility. The Viergever *et al* (2010) checklist of good practice was also identified.

Table 12 summarises how the methodology, process and methods were designed to fulfil the quality criteria. Table 13 demonstrates how the good practice checklist compares with the process in this chapter.

Process Quality Indicator	Aspect of process fulfilling quality indicator
Transparency	Clearly defined process
	Plan for description of analysis and with recorded areas of disagreement
	Reporting of reasons for prioritising ideas
Fairness/Inclusion	Designed as multi-stakeholder process
	Iterative development of stakeholder categories using multiple perspectives to minimise risk of bias in choice of stakeholders
	Consideration of strategies to avoid subtle exclusion:
	Purposive sampling in some methods
	Recruitment strategies to involve 'unorganised patients'
	Use of trained facilitators and PPI coordinator
	Inductive use of Framework analysis to develop shared meaning
Credibility	Triangulation through:
	Multiple sources used for stakeholder identification (literature review expert advice, consultation)
	Use of multiple methods in the process
	Use of different ways to record data within methods involving groups (transcripts, participant notes and field notes)
	Multiple researchers from different backgrounds analysing the data
	Checking results through the stages of analysis with stakeholders in the Steering Committee, Expert Forum and Consensus Conference
	Clearly recorded audit trail from original data to final priorities

Table 12 Quality indicators for the process

Viergever checklist	Consideration for this exercise
Preparatory work	
Determine context of exercise and resources available	Determined before process developed: outlined in Chapter 1
Use a comprehensive approach, either pre-set or own methods	Review of the literature used to develop an informed, comprehensive approach of own methods
Decide who should be included (inclusiveness)	Iterative approach to stakeholder identification using multiple sources
Choose what information should be gathered to inform the exercise	Choice determined by methodology used to answer research question. Stakeholder views to be gathered; current evidence to be considered in context through Expert Forum
Plan for implementation	Considered in choice of stakeholders participating in exercise; full implementation strategies outside scope of thesis (see further discussion in Chapter 6)
Decide priorities	
Select criteria for setting priorities: three dimensions – public health benefit, feasibility, cost	Methodological choice determines that criteria to be determined inductively from the stakeholders; plan for transparent reporting of these criteria
Choose methods for deciding priorities	Own methods chosen with rationale given based on the literature review
After priorities set	
Evaluate the priorities	Will form basis of discussion in thesis (will fall in scope of Chapter 6)
Write a clear report	Plan for clear reporting of findings and reasons for choices, with dissenting opinions included; put into context in Chapter 6; report to be given to stakeholders will be included in an Appendix (Appendix 14, page 361)

Table 13 Quality of exercise considered through the Viergever et al (2010) checklist

3.5 Data management and information governance

Provision was made throughout the protocol for information governance and data protection. All respondents to the survey were anonymous, and the responses participants in the focus groups were anonymised in transcripts. Participants in the Expert Forum and Consensus Conference were identified in appendices of the reports with consent, but no comments made were individually identifiable. Photography of group discussion sessions was done with consent and images were only used with signed consent from participants.

Survey responses were collected through Survey Monkey. Responses submitted on paper were entered into Survey Monkey and the paper copies were kept in a locked drawer on UCL premises. After the survey had closed the results were downloaded into an Excel spreadsheet and stored in a password-protected UCL database. All audio files were independently transcribed into a Word document by a UCL approved transcription service, except for the Expert Forum, which Researcher 1 (thesis author) transcribed due to poor sound quality. Researcher 1 proofread the transcriptions with the audio file to check for accuracy, identify any unclear words or terminology and clarify any areas of confusion. All electronic recordings and records were saved on a password-protected computer, backed up regularly and only shared within the Academic Team.

Each survey respondent was allocated a unique ID number based on the numbered line of the Excel spreadsheet in which they appeared. Any identifiable data from Survey Monkey, such as IP addresses, were deleted. Each focus group was allocated a unique focus group number, which was used to name audio files and transcript documents.

Microsoft Excel was used as the principle data management software, with Microsoft Word being used for written data reports and briefing documents. This was chosen pragmatically as it was familiar and accessible to all members of the Academic Team.

3.6 Ethics

Research priority setting is viewed as service evaluation and development by the NHS Research Ethics Service (NRES) (Cowan and Oliver 2013). This was reinforced by using the online Health Research Authority decision-making tool (HRA Decision Tool), which confirmed that this was not a study requiring ethical approval by NRES or the NHS Research and Development Office (NHS R&D). This study was further discussed with the UCL Research Ethics Committee, who agreed that it did not require formal ethical approval.

The following two chapters detail the findings of the research priority setting exercise for ENT, Hearing and Balance Care in the UK that has been described in this chapter.

Chapter 4: Findings: Stakeholders and the framework

4.1 Introduction

The previous chapter described the multi-stage process using multiple methods to determine the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders (see Figure 4, page 70). The analysis resulted in:

- A stakeholder map;

- A framework for the research priorities;

- A long list of research ideas reduced to a short list;

- Mapping of the proposed priorities back to the framework and stakeholder map;

- A final list of the research priorities;

- A description of the reasons why these priorities were chosen.

This chapter addresses the research question: "Who are the stakeholders for UK ENT, Hearing and Balance care?" It also begins to consider the research question: "What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?" This chapter describes the stakeholder map with the spread of stakeholders engaged in the exercise, and the framework.

4.2 The stakeholders

The stakeholders identified through the literature review and consultation with the Academic Team and Steering Committee were sorted into six categories (Figure 7). Table 14 provides details of the stakeholders in each category.
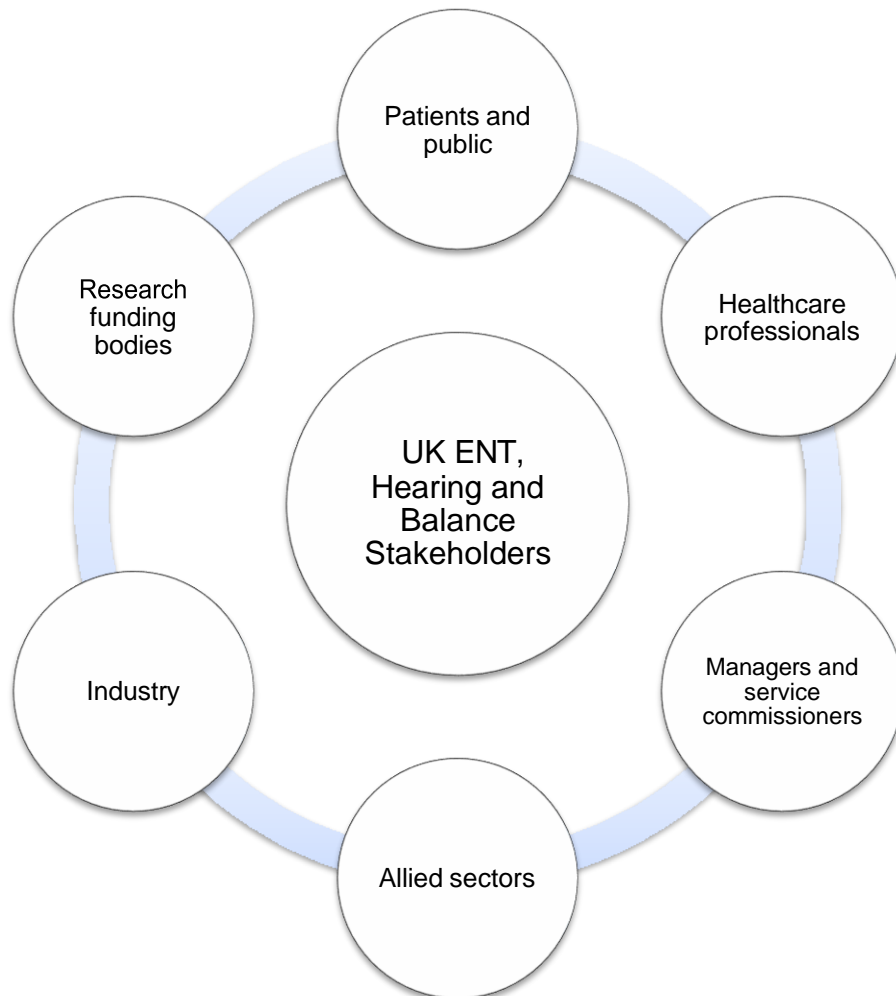
Figure 7 ENT, Hearing and Balance UK stakeholder categories

Stakeholder category	Descriptions of stakeholders approached
Patients and the Public	Charities and patient organisations for specific ENT, hearing and balance conditions Individual patients and carers through hospital clinics (London, Oxford, Belfast, Norwich) Public organisations without an affiliation to a specific patient group or condition Local patient and public involvement research groups
Healthcare Professionals	Specialty Royal Colleges Healthcare professional bodies NIHR Specialty Groups Individual clinicians through UK ENT department clinical directors/leads and through specialty conferences
Managers and Service Commissioners	Commissioning groups in all four nations Professional networks of healthcare managers Individual service managers through contacting clinical directors or leads of UK ENT departments
Allied Sectors	Professional organisations, e.g. Teachers of the Deaf Researchers and academics through academic units, e.g. NIHR Biomedical Research Units Policy-making bodies, e.g. NICE, NHS England
Industry	Biomedical companies specific to ENT, Hearing and Balance Biomedical companies without specialty-specific focus Private service providers
Research Funding Bodies	Government funded non-departmental public bodies, e.g. the NIHR Independent national charities without specific specialty remit Some overlap with charities specific to this field that fund research as well as represent patients, such as Action on Hearing Loss.

Table 14 Stakeholder categories

Figures 8 to 13 and tables 15 – 18 on pages 97 - 102 below present the details of the stakeholders for each method used in the prioritisation process:

Survey: There were 683 respondents to the survey. 48 of these (7% of all respondents) answered with random letters, numbers or words and were excluded from further analysis. The figures and tables on pages 97 - 99 show the range of respondents to the survey. As the survey was distributed through external organisations with private membership lists, social media and public events, data on the number of recipients of the survey are not available. Thus, an overall response rate to the survey could not be calculated.
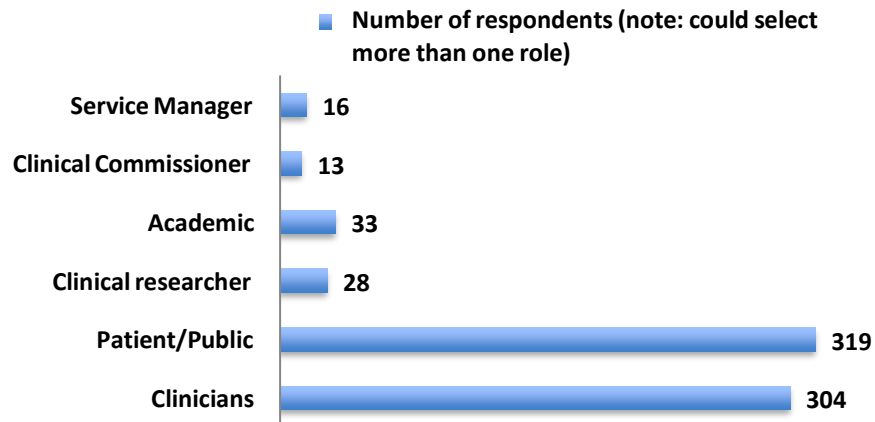
Figure 9 Survey respondents by stakeholder role
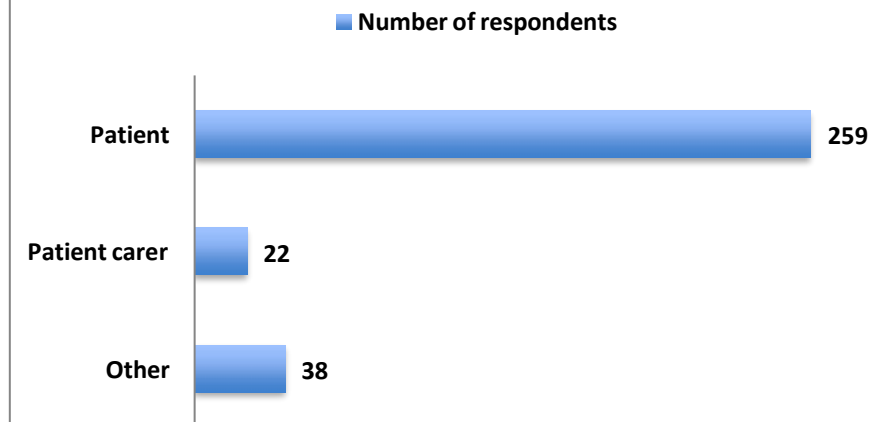
Figure 8 Patient and Public survey respondents
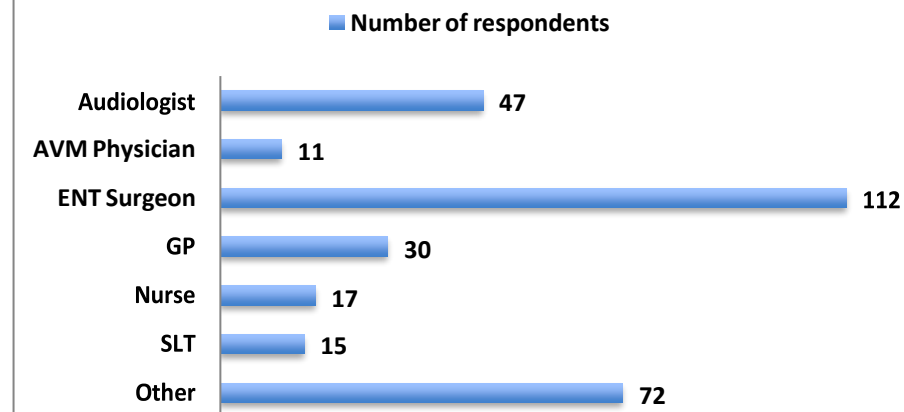
Figure 10 Clinician respondents to the survey

Specialty/Discipline	Number respondents	Specialty/Discipline	Number respondents
Paediatrician	9	Consultant Emergency Medicine	1
Hearing therapist	5	Educational audiologist	1
Community paediatrician	3	Histopathologist research assistant	1
Clinical scientist in audiology	3	Neurosurgeon	1
Neurologist	3	Operating Dept Practitioner	1
Geriatrician	3	Orthodontist	1
Anaesthetist	2	Pain Consultant	1
Psychologist	2	Pharmacist	1
Vestibular physiotherapist	2	Phlebotomist	1
Acoustician	1	Advanced Nurse Practitioner	1
Community ENT doctor	1	Students (PhD, medicine, nursing)	4

Table 15 Survey respondents from other clinical specialties or disciplines

Note: 72 respondents selected the "Other" option under clinician; 24 of these identified themselves as patients/patient carers rather than other clinical specialties or disciplines

Age range of respondents

■ Number of respondents

Age range	Number of respondents
80 or older	7
70-79	34
60-69	107
50-59	181
40-49	134
30-39	99
21-29	33
18-20	2
17 or younger	4

Number of respondents by country

■ Number of respondents

Country	Number of respondents
Wales	19
Scotland	52
Northern Ireland	16
England	492
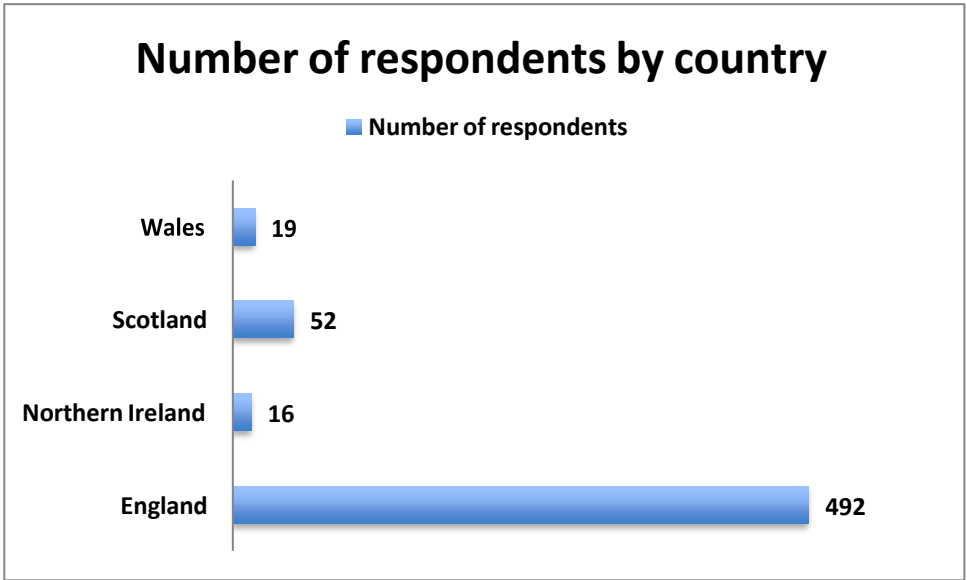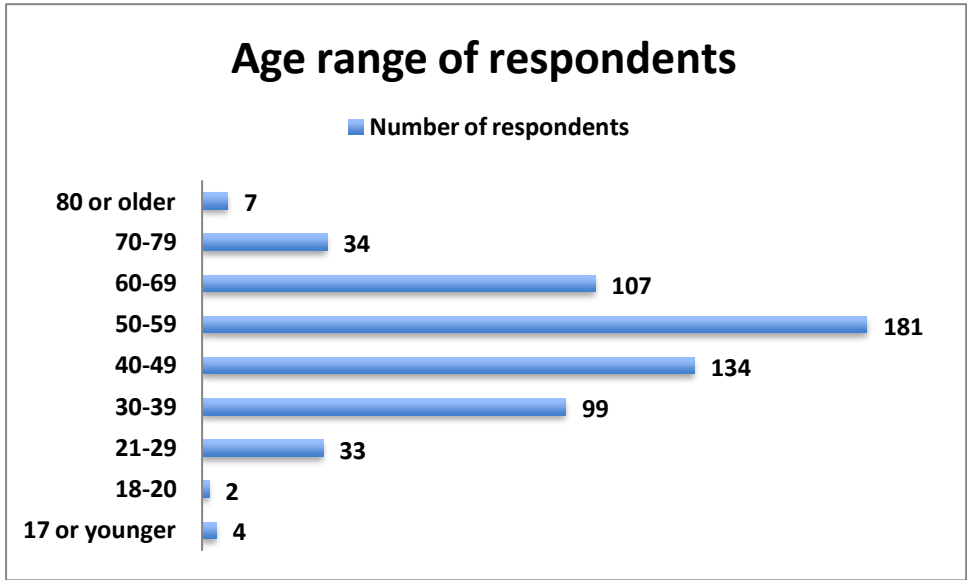
Figure 13 Age range of survey respondents Figure 12 Survey respondents by country

Ethnic Group of Respondents

■ Number of respondents

Ethnic group	Number of respondents
Other ethnic group	14
Mixed/multiple ethnic groups	10
Black/African/Caribbean/Black British	5
Asian/Asian British	38
White	483
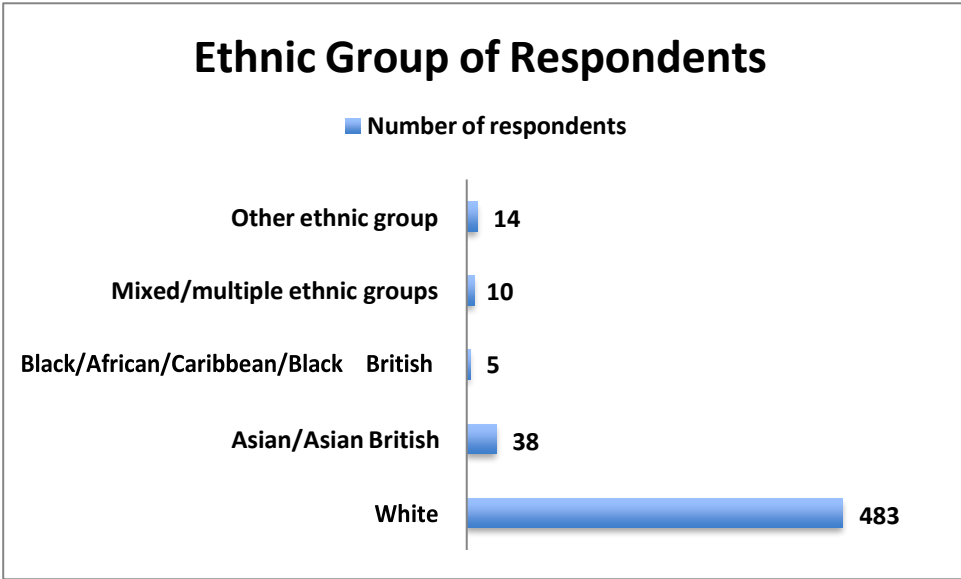
Figure 11 Survey respondents by ethnic group

Focus groups: Five focus groups were held. Information on age and self-declared ethnic origin was incomplete.

Focus Group	Participants	Details	Country	Age	Gender	Ethnicity
Healthcare professionals (FG 1)	9	Audiologists n=3 Paed audiologist n=1 ENT surgeon n=1 AV physician n=3 Hearing therapist n=1	England n=8 Wales n=1	All 40 – 59 years	Female n=6 Male n=3	White n=5 Asian/Br Asian n=2 Mixed n=1 Other n=1
Clinical academics + industry (FG 2)	16	Clinical academics n=10 Industry n=6 (Pharmaceuticals and Medical Tech/Devices)	England n=15 Scotland n=1	Majority did not disclose	Female n=5 Male n=11	Majority did not disclose
Patient and public (FG 3)	10	Ear/hearing/balance condition n=6 Benign throat condition n=2 Head and neck cancer n=1 Sinonasal condition n=1 Carer n=3 (carers also had conditions so n >10)	England n=9 Scotland n=1	40 – 49 n=2 50 – 59 n=2 60 – 69 n=4 70 – 79 n=1 did not state n=1	Female n=6 Male n=4	White n=8 Asian/Br Asian n=2
GPs + community based HCPs (FG 4)	4	GP n=1 GPSI/CCG member n=1 Community pharmacist n=1 Community nurse n=1	England n=4	30 – 39 n=2 40 – 49 n=1 50 – 59 n=1	Female n=2 Male n=2	White n=4
Commissioners + service managers (FG 5)	3	CCG member n=2 Hospital manager n=1	England n=3	30 – 39 n=1 40 – 49 n=1 50 – 59 n=1	Female n=2 Male n=1	White n=3

Table 16 Details and demographics of focus groups at data collection stage

Expert Forum: Table 17 shows the number and range of participants that were involved in the discussions that reduced the long list of research ideas to produce the shorter list for the Consensus Conference.

Meeting	Number participants	Stakeholders	Demographics
Expert Forum	21	15 clinical experts: 7 clinical professors ENT 2 ENT surgeons 1 audiovestibular physician 1 audiologist 2 clinical professors (general practice) 1 research nurse clinical lecturer 1 speech and language therapist *Three had roles in hospital management and governance; one was a clinical commissioner. Range of research interests from discovery science to translational, clinical and health services research, and across therapeutic areas.* 6 non-clinical experts: Expert patient representative (university PPI board; Hospital Board) 4 non-clinical professors (Epidemiology and Informatics; Innovation; Gerontology and Public Health; Health Services Research) NIHR Programme Director	All participants from England 7 female participants, 14 male participants. Age range and ethnic data were not collected.

Table 17 Expert Forum participants and demographics

Consensus Conference: The number and range of participants that determined the list of priorities from the short list of research ideas are shown in Table 18.

Meeting	Number participants	Stakeholders	Demographics
Consensus Conference	48	27 healthcare professionals (*additional roles – clinical academics, managers*): 15 ENT surgeons 4 nurses 2 GPs 2 paediatricians 2 audiologists 1 audiovestibular physician 1 respiratory physician 16 patients/carers: 11 adults (1 recruited from a charity; 2 from established patient participation groups; 8 from UCL PPI database with varying range of research experience) 3 young people (aged 21 years or below, part of an established PPI group) 2 parents of children with ENT conditions (recruited through local playgroups) 5 participants from research funders and allied professionals: NICE MRC and NIHR Action on Hearing Loss	Form completed by 22 of 48 participants: 21 from England; 1 from Northern Ireland 12 female; 10 male Age range from under 17 to 79 years old 16 White; 4 Asian/Asian British; 1 Black/African/Caribbean/Black British; 1 Mixed/multiple ethnic groups.

Table 18 Consensus Conference participants and demographics

4.3 The framework

4.3.1 The framework

The framework for the research priorities is shown in figure 14. This evolved as a two-dimensional matrix of interpretive themes and descriptive clinically oriented topics developed from the survey responses and focus group transcripts. These are explained and described in the following sections with iterations of their development. They reflect the need to establish a balance between expected recognisable clinical and research concepts and the more abstract concepts arising from the data.

4.3.2 The themes

Initial iterations: As described in Chapter 3 members of the Academic Team analysed the survey responses and focus group transcripts independently. Table 19 (pages 105 - 106) shows the initial themes developed by Researcher 1, the author of the thesis, with illustrative quotes from the survey. Figure 15 (page 107) shows the initial themes developed by other members of the Academic Team.

At this stage of the analysis there needed to be a clear focus on integrating and synthesising the themes, refining their definitions in order to develop the framework into which the raw data could be indexed, as per Ritchie, Spencer *et al* (2003). The independently developed themes were discussed by the Academic Team and refined to seven themes as shown in Table 20 (page 108).

Figure 14 The framework of themes and topics

Research Topics

| Inflammation, infection and allergy in adults | Head, neck and airway problems | Balance problems | Adult hearing loss and tinnitus | Children and young people | An Ageing Population |

Themes

Understanding health, illness and disease

Prevention of ill health, maintenance of good health

Clinical and cost-effectiveness

Designing and delivering health care

Creating effective communication

Promoting knowledge and learning

Theme	Ideas in theme	Illustrative quote
Participatory medicine/research	Patients as experts in their conditions; Patients working in partnership in their healthcare; Patients working in partnership in research; Empowering patients in this role.	"How to empower the researchers and clinicians involved with MdDS so that they can share their data - and their ideas - and access the acknowledged 'patient expertise'." (Resp 582)
Healthcare for the individual	Personalised medicine - targeting cures/therapies/management to the individual based on unique presentation/biological information [*personalised medicine beyond biomarkers*]; Managing individual care; Defining and maximising wellness for the individual.	"Approximately half the risk of developing any disease is due to genetic (genomic) factors yet we don't bother even attempting to consider making smarter genetically-informed diagnoses. Why ever not?" (Resp 602)
Maintaining function/prevention of disease	Stopping the onset of deterioration; Stopping or slowing the progression of deterioration; Maintaining wellness and function.	"Prevention of head and neck cancer… Head and neck cancer is increasing especially in the younger population." (Resp 123)
Holistic medicine/research	Systems or holistic approaches to studying and understanding disease; Impact of other factors such as ageing; Collaborative approaches; Considering the capacity needed for this approach; Global perspective – using evidence from other countries.	"Implantable aids (auditory and vestibular) are the biggest area of development in Otology in the last 30 years and our UK research profile in these areas has been minimal for many years. Interfacing with large Bio-engineering Universities seems important." (Resp 287)
Healthcare for the population	Considering burden of disease and common conditions; Economic impacts and cost effectiveness; Practice variation; The politics of healthcare.	"Any research into people with a learning disability who have eating drinking and swallowing problems - a great many… Little research is conducted with this client group. Health inequalities are greater. Access to health services is more challenging. Life expectancy is lower and mortality rates from asphyxiation and respiratory diseases are significantly higher." (Resp 155)

Technology and Innovation	Utilising new and existing technology in managing and delivering healthcare; The use of information technology; technology and ideas from other sectors; Disruptive ideas.	"Role of emerging communication technology in hearing (e.g. digital communication devices, self-fitting of hearing aids, Brain computer interfaces for controlling CIs and hearing aids)" (Resp 186)
Pathways	Development and design from presentation to final management; Barriers, access and unmet need; Timeliness of access/interventions/management; Safety and risk management; Place of management/treatment; capacity in pathways – human resources/equipment.	"Quicker diagnosis and liaising with primary healthcare providers i.e. GPs and practice nurses to aid the management of long term conditions like Menieres, tinnitus and hearing loss to enable patients easier access to help when they need it." (Resp 111)
Interventions	Conservative; Medical; Surgical; Alternative/non-medical.	"What are the best methods to assess and treat voice disorders?" (Resp 97)
Communication and education	Training and educating patients and healthcare professionals and promoting communication between them.	"support to the family should be readily offered and available - most families simply are not aware of the support available" (Resp 98)
Maintaining quality of life/minimising life altering impact	Managing long term conditions; Impact on life opportunities; Psychosocial impact.	"Does early diagnoses and management of hearing loss in aging population reduce risk of dementia? ; Aging population is increasing and so there are some concerns that undiagnosed hearing loss will increases cognitive decline and dementia- can early diagnoses reduce this risk?" (Resp 51)
Outcomes and increasing value	Research outcomes and appropriate measurements; Implementation; Impact on patient care.	"Accurate survival, functional and quality of life outcome measures for head and neck cancers to advise patients, clinicians and decision making" (Resp 46)

Table 19 Initial themes for Researcher 1

Themes for Researcher 2:

Consider the impact, barriers and facilitators of

- Cause
- Assessment
- Cure
- Prevention
- Assessing life satisfaction and new areas of evaluating quality of life

with an emphasis on all stakeholders taking responsibility for healthcare. The effect on aging /dementia and function, impact on family and social interaction, the accessibility of information and services, and the need for clear and better communication were noted throughout.

Themes for Researcher 3:

- "What works for me:" considering patients and their treatment on an individual level.
- Appreciating the patient voice.
- Patient participation in decision-making.
- Providing information to patients.
- Considering the broader impact of conditions, including the psychosocial impact.
- Collaboration across settings and in research: setting up registries and databases; using big data and utilising the data that already exists.
- Innovation
- Multimorbidity
- Holistic medicine
- Risk-based stratified medicine: considering biomarkers.
- Why do guidelines not work? This was especially noted in relation to OME ("*glue ear*").
- Improving pathways.
- Empowering GPs

Themes for Researcher 4:

- Prevention, such as in the area of age-related hearing loss
- The use of improved technology and its use in diagnosis, surgical technique and improved clinical practice
- Communication: improved communication between healthcare professionals and patients; information for patients; clinical information and its impact; information for clinicians and GPs.
- Understanding burden of disease in a wider context. This encompasses personal and societal burden of disease. It includes psychosocial burden of disease and appreciating its impact.
- Service delivery: encompassing redesign of delivery and patient pathways.
- Workforce training issues: this includes considering who should treat conditions and where treatment should take place with training happening accordingly.
- Self-management: helping patients to help themselves; encouraging patients to "not just put up with conditions" such as dizziness in the elderly.

Two further ideas around adherence to management/treatment and polypharmacy noted, but these could be incorporated into the larger themes with some consideration.

Themes for Researcher 5:

- Social burden of disease
- Service design
- Rare conditions versus common conditions
- Patient perceptions of clinical competence
- The capacity and capability of systems
- Patients as experts, often knowing more due to "internet expertise"
- The profile of research alongside service delivery
- The balance between needs of society and needs of the individual

Notes a distinction needed to be drawn between descriptive themes and interpretive themes.

Figure 15 Initial themes for Researchers 2 - 5

Maintaining functional ability	To encompass prevention and stopping deterioration, incorporating concepts such as "Cure" (Researcher 2) and "Considering patients and treatment on an individual level" (Researcher 3). Concept of a focus on functional ability, rather than on disease.
Service design and delivery	Agreed to more accurately reflect what emerged from the data. The concept of pathways conceptualised as part of a wider theme of service design and delivery. Theme to include: when and where service is delivered; barriers to access; pathways; considerations of integrated care; workforce considerations.
Communication	Highlighted independently by four of the five members of the Academic team as a separate theme due to repeated mention in the survey. It was agreed to keep this as a separate theme for further data analysis despite debate in the Academic Team as to whether it should be a part of the previous theme, Service Design and Delivery (Minutes Supervisory Meeting 08/12/2014; Minutes Supervisory Meeting 26/01/2015).
Education and training	Identified independently by three members of the Academic Team. Encompasses research to improve training and education of both healthcare professionals and patients, incorporating the theme of empowering the patient. Also includes the need for research to maximise human capacity in pathways, which was reinforced by the concepts of workforce training issues and the impact on capacity and capability of systems identified by two other team members.
Burden of Disease	Themes relating to understanding and assessing the burden of disease at an individual and population level identified by all Academic Team members, encompassing personal and societal aspects of disease, including psychosocial and quality of life impacts. Incorporates the concepts of impact on the individual; understanding the individual; and the burden of disease.
Using and developing technology	Uniting the themes of developing new technology and improving current technology, and combining my themes of interventions with technology and interventions. The definition of technology to be clarified with further discussion and analysis.
Innovations in research	An integration of ideas from three Academic Team members. Includes concepts of developing collaborations and different approaches to research, such as setting up registries and the use of big data, as well as increasing the profile of research alongside service delivery.

Table 20 First iteration combined themes

Development to final themes: Three of these initial themes developed so that their final iterations were similar to their initial definitions:

"Service design and delivery" evolved into into "Designing and delivering health care." Discussion and reference to the focus group transcripts considered whether these ideas were questions for research or if they were system design issues. The focus group discussions revolved around the scale of the problems. One of the participants in focus group 4 stated that: "…there's obviously big movements and worries and sort of drives and things happening on a very large scale about how to manage this and, as I say, it's very political, and I don't know whether as a research group maybe we'd be taking on too much. (Laughing)" (Focus Group 4 transcript, p 10: 476 – 479). This broad scope was echoed in Focus Group 5 where it was recognised that there was a move away for viewing simple service pathways to "more holistic" healthcare (Focus Group 5 transcript, p 11: 454 – 462). A further iteration of the initial themes based on ongoing analysis of the focus group discussions considered whether the ideas of patient-centred care and sustainability of health care should become part of the framework. After considering the discussion in the Expert Forum it was agreed that these potential themes represented a shift of focus from services to the wider issues of health care, which resulted in the final iteration of the theme.

"Education and training" and "Communication" were the subject of extended debate. Two issues arose with these original definitions. Firstly, was whether "Education" was an accurate representation of the concept developed from

the data; secondly was whether "Communication" was a theme in its own right.

The criticism of using "Education" as the title of the theme was that the survey responses and focus group discussions supported the expansion of this concept beyond a traditional understanding of training healthcare professionals. In the Expert Forum it was noted that "education" implied a teacher and a recipient, a formalised hierarchical structure. What had been described through the data went beyond this to considering different agents and ways to deliver knowledge and learning. It included providing patients opportunities to be able to learn and highlighted the potential for patient empowerment and self-management through learning, and especially through peer or group support. It also included the transfer of experiential knowledge from patient to clinician.

Participants in the Expert Forum further felt that "Education" did not adequately express the need to understand the barriers to changing behaviour through learning. There was concern that oversimplifying this would be a barrier to funding, as research funders do not fund education. Based on this feedback and on reference back to the data, it was agreed this theme would be titled "Promoting knowledge and learning."

Whether the theme of "Creating effective communication" should stand alone in its own right was a matter of debate throughout the analysis. When the theme of "Education" was redefined as "Promoting knowledge and learning" it

was asked if "Communication" was a subset of this broader theme. There was a considerable weight of response that indicated that a separate theme was appropriate. Survey respondents, most notably from the patient and public stakeholder category, highlighted the need for developing better interpersonal communication and the impact it had on health outcomes. Furthermore, the data indicated that the concept of helping patients to make treatment choices went beyond the simple sharing of information. Even with information, it was noted that patients may need to be supported in decision-making. What was central to this theme was the concept of helping patients to make a choice, which depends on effective communication between a patient and a healthcare professional where both share information and ideas about preferences.

The other four themes ("Burden of disease", "Maintaining functional ability", "Using and developing technology" and "Innovations in research") changed significantly. This was partly because ideas around function overlapped with the other themes. For example, questions around the impact of ENT conditions on the individual and society were raised throughout the survey and through the focus groups. These were classified initially under the theme "Burden of disease." In different focus groups it was noted that there was a need for more qualitative research to understand the impact of these conditions on patients. Patient stakeholders in particular felt that the depth of suffering and impact on quality of life were underestimated, and their impact on daily function were not considered along with their ability to work and interact with society. This overlapped with "Maintaining functional ability," and

the idea of research to understand the impact of conditions on the function of individuals, their families and carers became part of this theme.

This evolving theme had the concept of 'understanding' at its core. From understanding the personal and societal impact of illness, the scope expanded to also understanding more about disease. As one survey respondent outlined: "…The link between bruxism, laryngopharyngeal reflux, stress and globus pharyngeus needs further research. The causes of the condition need to be determined with confidence. Treatment guidelines need to be issued to reduce unnecessary investigations and to provide patients with a useful treatment plan. To continue to claim, as a specialty, that the aetiology is 'unknown', and to inform patients that 'nothing is seriously wrong', does not address the patients concerns or allow for any sensible treatment plans. Massive burden of disease/illness in patients that receive no useful diagnosis or treatment from primary or secondary care…" (survey respondent 124).

This response summarises the extent of understanding needed: increasing knowledge of the causes and epidemiology of disease go on to influence how they are managed, and consequently how they affect people. The need for greater understanding of disease was echoed across focus groups and was a topic for discussion in four of the five groups. Thus, in considering 'understanding' being the core of the definition of this theme, it was expanded to include understanding disease, as well as its impact on the subjective experience of patients. This became the theme of "Understanding health, illness and disease."

With the concepts of function being incorporated into this newly defined theme, "Maintaining functional ability" was reconsidered. In its original definition this theme had also encompassed prevention (see table 20). Based on the weight of response across stakeholder categories in the survey, and on the recommendation of the Expert Forum, prevention strategies and interventions to maintain the function of individuals and the health and wellbeing across the population was established as a separate theme.

Finally, with understanding and preventative strategies removed from the initial theme of maintaining functional ability, what remained was the idea of treatments to restore or replace function. This overlapped with the theme of using and developing technology in relation to diagnosis, treatment and management. The question then became whether to define this around functionality. The Academic Team discussed whether this was too general and whether it would be unclear to the stakeholders. Instead, the concepts of clinical and cost effectiveness were suggested. Clinical effectiveness was suggested as it incorporated the concept of maintaining or improving patient function. Cost effectiveness was also identified as being a component of assessing diagnosis, treatment and management in the data. This was how the theme of "Clinical and cost effectiveness" was developed.

The final six themes for the framework with definitions and illustrative quotes are listed in table 21 below.

A further theme, "Innovations in research" had also been identified in the earlier iteration of the themes (Table 20). This was re-discussed by the Academic team. Despite a lack of prompts in the survey or focus group topic guides asking how to develop research, a spread of stakeholders proposed ideas about this. These ideas were not potential research questions, but rather were strategies for implementing the priorities and developing research projects. It was agreed that this theme would support the framework and priorities as implementation strategies for developing the research priorities into projects.

The specific ideas identified were:

- Increasing the quality and value of research performed. For example, one survey respondent wanted "More RCTs in common ENT conditions e.g. glue ear, rhinosinusitis, hearing rehabilitation. Relative lack of RCTs currently" (survey respondent 607).

- Developing research collaborations. This involved developing and facilitating networks and professional relationships both within the ENT, Hearing and Balance specialties as well as with other groups. Patients were recognised as key collaborators in research. This was also seen as a potential way of developing disruptive ideas (Focus Group 2 transcript).

- Networking platforms to support and facilitate research collaboration: "The network almost needs an interface where we contact every ENT surgeon and say, "If there were a trial, would you be willing to participate? What is the most common symptom or diagnosis that you see in your clinic?" And then from your own data you could log on that portal the number of

patients that come through your practice in a year, so that when industry want a trial they look and say, "Right, we need 200 patients; I can get them all in one area or if I want them spread over the country," they can immediately identify where to go and then it would facilitate, as you say, that partnership." (Focus Group 2 transcript, p 20: 841 – 849).

- Developing the capacity to perform the proposed research, and the capability of those who wished to engage in it. There was a particular focus on the need to support a new generation of researchers: "… I dearly wish there was someone, or some body or institution, which might offer support and encouragement to junior doctors who have original ideas and sufficient motivation to carry their ideas through to completion …" (survey respondent 290).

- Setting up data collection platforms, such condition-specific registries collecting real world data: "In the 21st century we still have no idea about such basic, common issues. Perhaps there should be national databases for such cases (e.g. the acute SNHL group) to track epidemiology, natural history and treatment modalities" (survey respondent 90).

Theme	Definition	Example of responses leading to theme
Understanding health, illness and disease	Developing an understanding of ENT, Hearing and Balance conditions, and the impact they have on health and illness in individuals, their families and society	"...actually are we at the stage of talking about treatments? Shouldn't we focus more on aetiology, causation and these are areas that may not come up…" (Focus group 2 participant). "How reduced hearing affects quality of life. How tinnitus affects quality of life. How poor balance affects quality of life. Because poor hearing, tinnitus and poor balance can leave a person feeling isolated, which can in turn affect their relationships, job prospects and mental health." (Survey respondent 417)
Prevention of ill health, maintenance of good health	Strategies and interventions that prevent, stop or slow loss of function, and that maintain health and wellbeing across the population	"And what about the role of public health? If EVERYONE did a little bit of exercise, ate properly and didn't get fat, and didn't smoke, a huge health care burden would be lifted without having to do anything else. Because we only have a limited pot for state-delivered health care and we had better start working out what we should be spending our money on to make it go furthest. I say we should start by looking at public health measures and then look at the relative values of the contribution of various clinical specialities" (Survey respondent 36)
Clinical and cost-effectiveness	Testing existing and new ways to diagnose, treat and manage patients and their "conditions"	"What is the long-term impact of using a hearing aid? Are there any adverse issues related to delay or non-provision of amplification until a severe hearing loss it reached?... The above questions are important as we approach an era of even greater austerity and an ever increasing elderly population" (Survey respondent 472)

Designing and delivering health care	Strategies and interventions for improving the quality and value of health care, and making it sustainable and patient-centred	"…there are so many areas, not only from the technological side but also from beyond the technological side how to develop the services, how to run the services which can be looked into purely from a research point of view. But it doesn't get discussed very often that is the problem." (Focus Group 1 participant)
Creating effective communication	Improving health and services through better communication and interpersonal interactions	"Meniere's Disease!!! I have read HORROR stories of the unsympathetic and insensitive treatment of patients with Meniere's Disease and could add a few bad experiences as well. Please do research on the way GPs and ENT's recognise and treat their patients with MD so their empathy and understanding of the psychological effects their approach may have on patients can be improved" (survey respondent 127)
Promoting knowledge and learning	Interventions and strategies to improve health, function and service through knowledge sharing and learning for patients, healthcare professionals and providers.	"Paediatric balance disorder – similarly, under diagnosed due to lack of knowledge and enthusiasm. Mostly ignored as the symptoms are not overt and lack specialist vestibular rehabilitation studies (services). Developmental delay, cyclical vomiting, (abdominal) migraine, travel sickness all directly related to vestibular cause and potentially curable with targeted vestibular rehab" (Survey respondent 674)

Table 21 Themes, definitions and illustrative quote

4.3.3 The topics

Through the group analysis it was noted that there were a combination of descriptive clinical topics and interpretive themes identified from the data. This was highlighted in the original analysis by Researcher 5 (see Figure 15). The clinical academics in the team were quick to identify clinical topics that spanned the data. This reflected their experience and it was acknowledged that some of the stakeholders would expect to see these in the outputs of the exercise. The Academic team had extensive discussions about whether there was a need to recognise conditions as part of the framework or whether this was an imposed biomedical approach that did not reflect the voices of the stakeholders. It was suggested that the framework should become a matrix of interpretive themes and clinical topics drawn from the responses. This represented a pragmatic balance to not alienate stakeholders such as clinicians, clinical researchers and research funders. Furthermore, it was recognised that patient stakeholders framed their responses to the survey and focus group discussions around their personal experience of their diagnosed condition. Thus, what could be viewed as a biomedical approach was inherent in patient stakeholder responses. This tension between remaining faithful to a purely interpretive, inductive approach, and the results being familiar to the stakeholders, was revisited several times during the analysis.

On charting the data in the themes, it became apparent that developing topics on individual conditions, such as Ménière's Disease or paediatric allergic rhinosinusitis would be too granular. This decision was made for two reasons. Firstly, a higher-level analysis with broader topics that incorporated conditions, symptoms, problems

and patient characteristics was felt to be a more faithful interpretation of the stakeholder responses. This again represented a balance between the tensions outlined above. Secondly, this was a pragmatic decision as there were a very large number of individual conditions in the responses. These topics, with descriptions and illustrative quotes are summarised in Figure 16.

Adult Hearing Loss and Tinnitus

"…In silence tinnitus is always there to remind you something's wrong, and in noisy environments the deafness is always there to remind you something is wrong, and that's why it's incredibly insidious" (Patient focus group participant).

Survey respondents overwhelmingly felt that more research was needed in all aspects of hearing loss and tinnitus and in all age groups. Hearing loss and tinnitus in children and older adults is dealt with under the relevant topics, although there is some overlap between these. Respondents also recognised the need for more tinnitus research, and were clear about the suffering that people with this condition experienced.

Balance Problems

"Balance disorders can have a catastrophic effect on the sufferer physically; emotionally; socially and in work settings. So continuing the promising research into causes and effective treatment are vital." (Survey respondent 371)

Survey respondents raised many questions about conditions affecting balance in all age groups. Our respondents felt it was under-researched and poorly understood, despite its significant impact on quality of life and ability to function on a daily basis. As one survey respondent noted when answering why this should be a priority: "You wouldn't ask if you had it."

An Ageing Population

"Well I think people are waking up to the big ticket items which is we're all getting older and, as we are fitter until we get older, that's only postponing the avalanche and people's expectations are still going up…" (Commissioner focus group participant).

Survey respondents recognised that an ageing population faced significant health challenges, often had multiple problems, and that ENT, Hearing and Balance issues could have a significant impact on them, their families and healthcare costs to society.

Inflammation, Infection and Allergy in Adults

"It's not life threatening. It's not visible. However smell and taste are TWO of the senses. This has a huge effect on the quality of life on a day to day, hour by hour, minute to minute basis" (Survey respondent 53).

This topic asks questions about inflammation and infections of the ear, nose and throat, and allergic conditions of the nose and sinuses. There were many questions about allergies. There is already research happening in these areas, with many questions being dealt with by specialists such as immunologists. ENT infections are very common in primary and secondary care, and affect adults and children. Some questions raised overlap with Children and Young People. These questions were put in the most appropriate topic.

Head, Neck and Airway

"Massive burden of disease/illness in patients that receive no useful diagnosis or treatment from primary or secondary care. Massive expenses to the NHS in repeat appointments and recurrent investigations, without a conclusion every being reached" (Survey respondent 124, talking about globus pharyngeus).

This deals with ENT conditions that affect voice, swallowing and breathing. GENERATE respondents had questions about Head and Neck cancer, but many of these are already being researched. There were also many questions about benign disease asked by our respondents. This increasingly accounts for time taken in clinic, and many clinicians are uncertain about the best ways of managing these conditions.

Children and Young People

"These are all very common conditions of childhood which have a significant impact on the child a short and long term as well as their educational attainment." (Survey respondent 58).

Survey respondents were concerned about the impact of ENT, hearing and balance conditions on children and young people, who are "…yet to live their life" (Survey respondent 49). Children at all ages were considered, as well as children with complex needs, who often have ENT problems.

Figure 16 The topics

120

The choice of language in developing the topics was important. Different stakeholders used different descriptions, ranging from technical/clinical terms to descriptive symptom-based wording. This was especially noticeable in research ideas relating to sinonasal conditions. How these were expressed differed considerably across the stakeholder groups, making it more challenging to define a topic area for these problems. The term "sinonasal" was a very specific ENT term. Patient stakeholders framed this in terms of physical manifestations such as lack of sense of smell, nasal blockage, or symptoms of allergy. Clinically or academically orientated stakeholders used clinical terms such as sinusitis, rhinosinusitis (Focus Group 4, p 15), or chronic inflammation (Focus Group 2, p11).

The difficulty in resolving these two different perspectives was discussed in the Expert Forum, where the conversation focused around the topic of anosmia. It was acknowledged that there was a significant weight of response around this. However, this was considered a symptom, not a condition, and therefore the focus was on inflammatory, infectious or allergic conditions that led to this symptom (Expert Forum Report, Appendix 10, p 288). This became the title of the topic. This had the advantage of not focusing on the granularity of individual symptoms, but was an adoption of more technically focused language than was used by many of the stakeholders. Nevertheless, it was felt to be a pragmatic, strategic choice in developing a framework that would resonate with research funders.

This problem in this area contrasted with the responses around ear problems, where terms such as hearing loss, deafness, tinnitus and balance were used more universally across the stakeholder categories. In order to reflect this language topics relating to these problems were described as "Hearing loss and tinnitus" and "Balance problems" rather than "Otology." A few of the ear problems described in the data did relate specifically to infection and inflammation, and these were included under that topic heading.

The problems of hearing and balance functions were both seen repeatedly across the themes and were suggested by survey respondents across the stakeholder categories. The question of whether to combine these as "Ear problems" was considered. However, the stand-alone importance of balance problems was emphasised in different focus groups as well as appearing separately in the survey responses. There were no dissenting opinions about this across the data. With this weight of opinion from respondents and focus group participants, "Balance Problems" became a separate topic from "Hearing Loss and Tinnitus".

The final set of conditions that could be identified across the thematic charts were those relating to cancers and benign problems of the head, neck and airway. These were described both in terms of functional problems, such as problems with breathing or snoring, voice and swallow, as well as specific conditions or disease processes, such as thyroid carcinoma or subglottic stenosis. Most of these responses could be easily conceptualised as part of a "Head, Neck and Airway" topic. However, in a similar manner to the ear

problems, some had an added dimension, such as pain or infection, which overlapped with the "Infection, Inflammation and Allergy" topic. These were conditions such as tonsillitis or general upper respiratory tract infections (URTIs), which were included with that section.

On analysis, what also appeared across the themes was the need for research in different populations of patients, namely children and the elderly, as separate from adults. Specific questions concerning these groups appeared frequently in each topic as well, justifying their consideration as separate topics in their own right.

This was an emotive topic for some of the survey respondents, who felt that children should be managed and considered in a different manner to adults:

"There doesn't seem to be much weight given to the life stage of hearing loss .. A hearing loss from birth has ALOT more affect than a mild age related loss from 50 on. Both in terms of impairment duration and 'importance'. E.g. Childhood / Education and employment are affected if hearing impaired from birth. Hearing loss experienced at 50 on is post most of the above events. Yet - ENT handles HI at younger ages the same as OAPs." (survey respondent 69).

The development of "Children and Young People" as a separate topic was supported by the spread of responses across the themes and stakeholder categories.

In a similar manner to the responses about children, there were responses across the themes and stakeholder categories that identified the elderly or ageing population as separate from the rest of the adult population. As stated by one of the respondents to the survey, what he felt was important were:

"Questions related to common conditions and procedures plus ageing population, co-morbidities, mental health problems e.g. dementia. So: dizziness, deafness, rhinosinusitis, globus, qol esp in cancer Rx." (survey respondent 684).

The Expert Forum noted this to be a heterogenous and changing population. It was suggested that rather than focus on age, the consideration should be on polypharmacy and multimorbidity (Expert Forum Report, Appendix 10, p288). This mirrored some of the discussion in Focus group 4. However, the weight of response in the data focused on age and a shifting demographic. Therefore, the topic was titled "An Ageing Population.". This reflected the language from the stakeholders and acknowledged the perception of problems with age highlighted above.

A representation of the development of these topics from the ideas from the survey and focus groups is displayed in Figures 17 and 18.

Figure 17 Development of condition-based topics

Vertigo/Dizziness:
Balance problem
Ménière's Disease
BPPV
MdDS
Labyrinthitis
Vestibular neuronitis
Falls in the elderly
Motion sickness
Vestibular problems:
In adult and elderly
In child

Ear congestion
Glue ear/OME/associated with
Down's or cleft palate
Eustachian tube dysfunction
Ear infections:
in adult and child
middle ear or external ear
Mastoiditis
SOM/CSOM
Cholesteatoma
Tympanic membrane
retraction
Ear eczema

In children, adults

Balance problems

Overlap with hearing issues

Hearing loss, tinnitus

Overlap with hearing issues

Features of pain, unwellness, discharge, fluid

Infection, Inflammation, Allergy

and the elderly

Hearing loss/Deafness:
Age-related
Noise induced
Hearing loss of childhood
Single-sided
Sudden sensorineural
Sensorineural (adult and child)
Conductive
APD and ANSD (adult and child)
Ototoxicity

Tinnitus:
In adult
In child

Acoustic Neuroma
Barotrauma (also features of pain)

Figure 18 Further development of "condition-based" topics

Head and neck cancer
Thyroid cancer
Subglottic stenosis
Laryngomalacia
Snoring
OSA (adult and child)
Voice loss/dysphonia/dystonia
Dysphagia
Globus pharyngeus
Speech and swallow disorders
Tongue tie and feeding
Cleft lip
Velopharyngeal inadequacy
LPR/chronic cough

Glue ear/OME/associated with
Down's or cleft palate
Eustachian tube dysfunction
Barotrauma
Ear infections:
in adult and child
middle ear or external ear
Mastoiditis
SOM/CSOM
Cholesteatoma
Tympanic membrane
retraction
Ear eczema

In children, adults

Head, Neck and Airway

Structural problems
Airway problems

Features of pain, unwellness, discharge, fluid

Infection, Inflammation, Allergy

and the elderly

Problems with the nose, nasal breathing and taste:
Anosmia (congenital and acquired
Phantosmia/cacosmia/parosmia
Reduced or absent taste sensation
Stuffed nose/nasal congestion or blockage
Nasal deformity
Epistaxis
Post-nasal drip (*chronic cough/LPR*)
Facial pain
Hayfever/allergies
Allergic rhinitis (adult and child)
Rhinosinusitis (acute, chronic, fungal)
Nasal polyps

URTIs (adult and child)/ENT infections
Tonsillitis
Sore throat

This chapter has presented the stakeholder map and the framework, which first part of the analysis of the research priorities of UK ENT, Hearing and Balance stakeholders. The next chapter gives the rest of that analysis with the map of the priorities to the framework and stakeholders, the final priorities and reasons for prioritising them.

Chapter 5: The priorities and criteria for prioritisation

5.1 Introduction

This chapter shows the development of the long list of research ideas and how this became the short list following the Expert Forum. It then gives the priorities agreed by the Consensus Conference, with the reasons for prioritisation. It also shows how the short list of ideas and agreed priorities map across the original data, stakeholder categories and framework. This chapter, combined with the results of the framework outlined in the previous one, answers the research question asked in this thesis: "What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?" It also addresses the final research question posed: "Why did the stakeholders prioritise those ideas?"

5.2 The research ideas

The survey responses and focus group transcripts gave rise to 1697 research suggestions. These were developed into research ideas, then refined and prioritised into 21 research priorities. Figure 19 shows how the number of ideas were reduced through the prioritisation process. Figure 20 shows the agreed priorities under the topic headings developed in the framework. The sections following the diagrams show how the ideas were developed to produce a long list, then a shorter list of ideas, and then discussed in the Consensus Conference to determine the research priorities.

Figure 19 Flowchart of ideas to priorities

1697 research ideas from the data

1027 duplicate/similar ideas

670 ideas for Expert Forum

572 ideas being addressed or ideas combined

98 ideas for Consensus Conference

77 ideas not prioritised

21 research priorities for Agenda

Figure 20 The agreed priorities by topic

Balance Problems:

- What is the optimum pathway for people with balance problems?
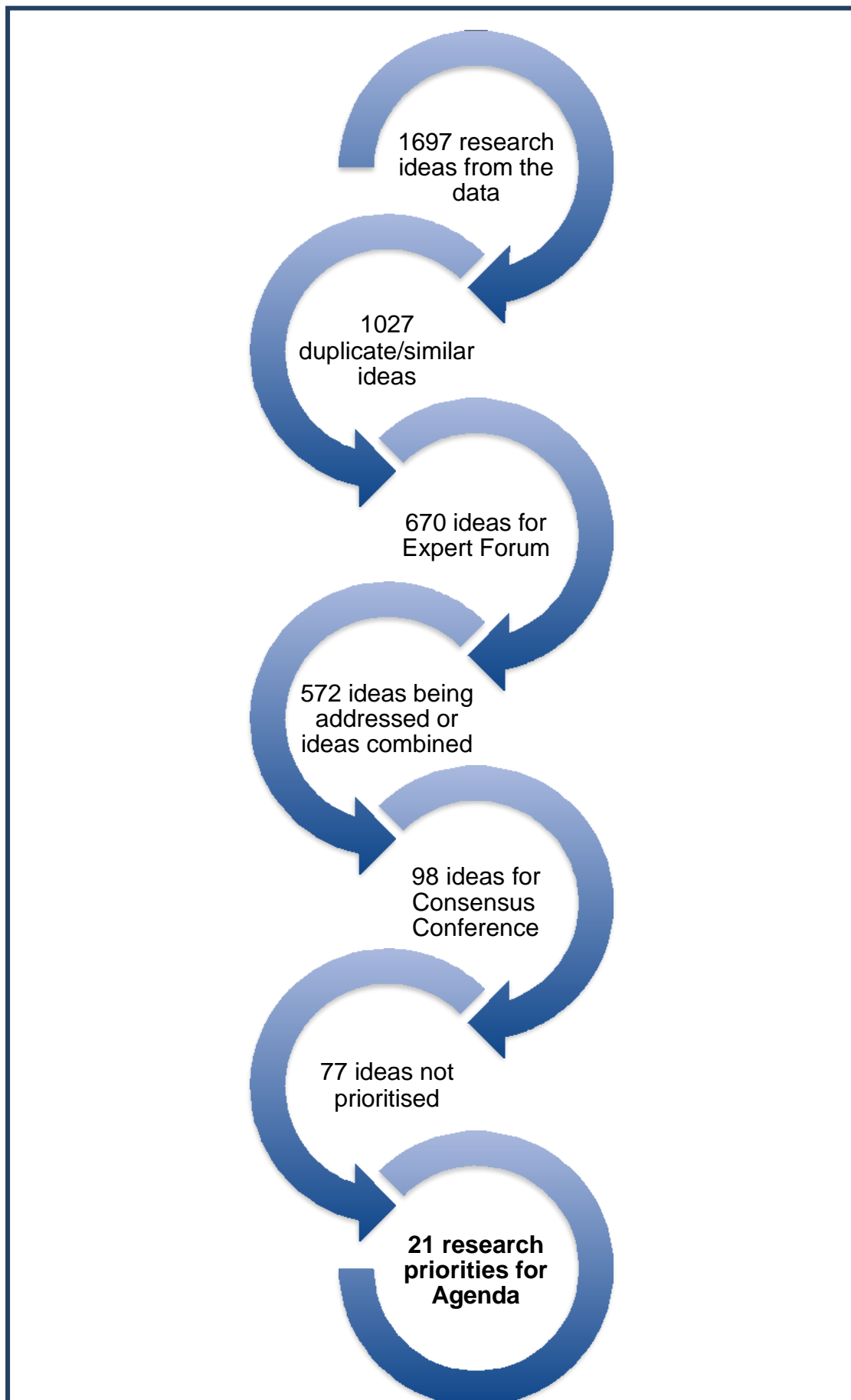   - What can be dealt with in primary care and the community, and what are the indications for referral?
    - How should specialist clinics be set up?
- How can patients be empowered to self-manage their balance problems and how does this impact on their outcomes?
- Develop clear diagnostic criteria for different balance disorders so that patients can be informed about prognosis and best treatment.

Head, Neck and Airway:

- What is the optimal management for adults with OSA?
- What outcome measures and instruments should we use for research into head, neck and airway disease, both benign and malignant?
- What is the role for improved diagnostics in head, neck and airway conditions?
- What is the optimal management for adults with voice problems?

Inflammation, Infection and Allergy in Adults:

- Define the different variations of disease that make up rhinosinusitis ("endotypes" and "phenotypes") and develop ways of classifying them based on signs, symptoms and biochemical markers.
- What are the most clinical and cost effective treatments for chronic rhinosinusitis?
- How do we improve the implementation of guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden and its impact on antibiotic resistance in adults and children?
- What outcome measures and instruments should we use in primary and secondary care for chronic ENT infections and inflammatory conditions?

Adult Hearing Loss and Tinnitus:

- What are the best management strategies for tinnitus?
- Which hearing aid strategies work best for adults with hearing loss?
- What is the burden and impact of hearing loss on an individual, family and population level?
- What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?

An Ageing Population:

- What are effective interventions for hearing loss in older people?
- Is it beneficial to introduce an adult hearing screening programme and how do we best develop it?
- What is the optimum design for "balance pathways" in older people with balance problems?
    - How and where can older people with balance problems and with multiple medical conditions and decreased mobility best be managed?
    - How can self-management strategies be built into these pathways and what is their impact on patient outcomes?

Children and Young People:

- What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?
- How do we best diagnose and manage obstructive sleep apnoea (OSA) in children?
- How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professionals in different settings, and what is the impact of this on patient outcome?

5.2.1 Developing the long list of research ideas

The survey and focus groups asked respondents and participants to suggest important "topics, questions or issues" for research, rather than defined research questions. This approach was chosen as it was more suitable for the range of stakeholders involved in the exercise. As a result of this broad approach the level of detail and how the ideas were expressed was highly variable. Some were detailed, technical ideas. Others were expressed as broader thoughts or statements, and some of the suggestions were relayed as shared experiences or areas of dissatisfaction, from which research ideas were derived. An example of the latter was survey respondent 68:

"Vestibular/Vertigo-related matters are of a personal concern. Sudden onset apparent BPPV followed by a misdiagnosis of Meniere's Disease with eventual diagnosis of Vestibular Migraine.   Quick, accurate diagnosis by NHS specialists - not private consultations. Link between vertigo and neck arthritis? Trapped nerve? I suffer both arthritis of the neck and 'vestibular migraine' (which I think is a catch-all for all unexplained vertiginous attacks). The vertigo first displayed itself very suddenly three years ago, following an investigated (privately) but undiagnosed pain in the side of the head which got worse on movement not on touch. Then a sharp gust of wind into the ear, followed within days by severe dizzy spells on movement of the head.   Again, privately, I was treated for BPPV and this seemed to work. But then a couple of months later, I was very dizzy and very sick. I was due an appt but this was some weeks ahead and it was not possible to bring it forward on the NHS. I had to wait. In the winter of 2011 I had such severe vertigo that I was knocked off my feet and it took two weeks to learn to walk again once I'd stopped being sick. Investigations were undertaken by local NHS ENT who initially suggested Meniere's Disease and this caused a stir at work where I was no longer allowed to park in the company car park... and other discriminatory behaviour. After hot and cold water in the ears and other gross tests I was told that I had vestibular migraine and discharged... No follow up. No progress check. No emergency number should I get a sudden attack...   I live in dread of it happening all over again."

Only one actual question was posed (link between vertigo and neck arthritis and trapped nerve) but clearly other ideas can be drawn from it. The response

came under the "Balance Problems" topic in the framework and contributed ideas across the themes (see figure 21).

One of the ideas from this response was the need to develop quick and accurate ways of diagnosing balance problems that are acceptable to patients. This was grouped with other similar responses such as:

"Balance Testing. More research on balance issues" (survey respondent 110).

"balance - alternatives to caloric test that are less invasive and give more ecologically valid data - vHIT goes someway towards this" (survey respondent 121)

"Vertigo- best ix and management" (survey respondent 338).

This resulted in a single research idea in the Clinical and Cost-Effectiveness theme:

"What is the effectiveness of early, correct diagnosis on outcome (quality of life/mental health) in patients with vestibular dysfunction? Develop more sensitive tests for diagnosis and consider less invasive alternatives to caloric testing (such as vHIT, VEMPS)."

This example demonstrates two points:

- a single response could give rise to multiple ideas,

- and similar ideas from different respondents were combined to give a single overarching research idea.

A table showing the final priorities and examples of data from which they were derived may be found in Appendix 13, page 356. The process of elimination

and combination of the ideas through the prioritisation process is described in greater detail in the following sections.
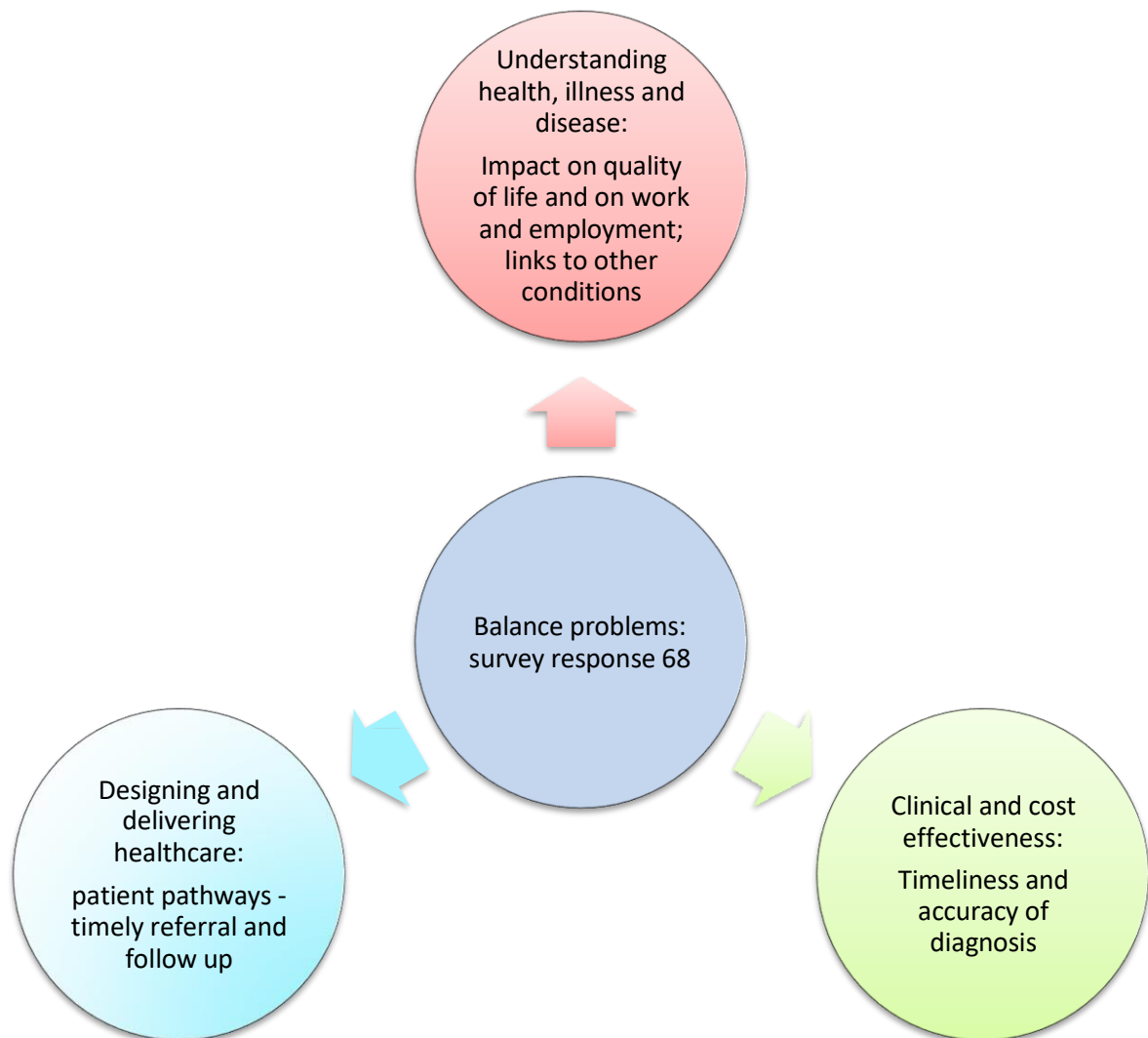
Figure 21 Ideas from survey respondent 68

5.2.2 Developing the short list of research ideas

The 670 research ideas taken to the Expert Forum were still a mixture of broad and granular ideas. In the small group discussions, the experts independently chose to distil the ideas so that several different but related ones were combined into broader overviews. There was not complete

concurrence with this approach however, as some participants felt that there was a risk of too great distillation. One expert noted that research funders might be looking for more specific research questions to fund. These two approaches were discussed by the Academic Team. As the approach of combining related ideas had arisen independently in the different small group discussions, and as the exercise was not designed to develop specific research questions, the Academic Team agreed that the long list ideas could be combined as per the experts' suggestions. This was further discussed with the Steering Committee, who agreed with this approach. Based on these discussions and using the expert recommendations, the ideas already addressed by research were excluded, and the other ideas were combined. A summary of the key points of discussion in the Expert Forum may be seen in table 22.

One of the concerns expressed by both the Steering Committee and the Expert Forum itself was that the use of experts rather than literature reviews would introduce the bias of the individual experts. The Expert Forum suggested that a mapping process be used from the original ideas and stakeholders to the agreed priorities so that there was accountability and an evidence trail. To assess for personal bias at this stage, the short list of research ideas was mapped across the framework developed by the Academic Team from the original survey and focus group data. Mapping also occurred after the Consensus Conference agreed priorities (see pages 147 - 151 below). The results of mapping the short list of research ideas are given in Table 23 on page 138 below.

Main points	Eliminated and combined ideas
Adult hearing loss and tinnitus Distillation of ideas with some points combined into broader overviews, with concern it was too distilled. Distilling involved: thinking at individual and population level; challenging preconceived ideas of what works, such as HAs; capturing "blue sky thinking," hence the inclusion of topics involving developments in molecular therapy. Is the accepted evidence up to date? Practice in this area and hearing aids have changed a lot since the evidence was developed. In this topic as well as the others we need to consider how to ensure that the evidence stays relevant. Designing healthcare: Optimal <u>system</u> – this considers designing a whole system that identifies and then manages those that would or may benefit from HAs/audition/tinnitus therapy. It was questioned if work around whether hearing loss is a predictor of Alzheimer's, and who develops hearing loss had been done and if this should be added.	Experts' focus was on combining questions as opposed to eliminating ideas. Multiple questions through all themes were combined by not referencing individual conditions, but rather the broader areas of hearing loss or tinnitus. Exceptions to the above were the retention of specific topic questions about management of single-sided deafness, acoustic neuroma, both of which were felt to be under represented in current research, and mild-moderate hearing loss which was noted to represent a significant burden of disease where current interventions were under-utilised. Questions about tertiary prevention/rehabilitation appeared to have been eliminated but were actually combined into Clinical and Cost-effectiveness questions about interventions. Screening questions were removed from this topic and combined into a question in An Ageing Population where they were felt to be more relevant.
Balance problems Experts concerned their assessment of this topic was not as thorough given its recognised real importance. Impact of locomotion and balance on individuals, families and society and the healthcare burden of older people falling were noted as <u>substantial</u>. Consideration given to population-based services – who are the people who are and aren't patients? Are we meeting their need? Get rid of primary/secondary care divide. Multi-morbidity noted to overlap with ageing and balance. Research capacity issue in finding people interested and capable in this topic identified. Research culture in balance research needs development; Audiovestibular community noted to be more advanced, but more cross-disciplinary work and more capacity needed.	Experts focused on combining ideas into broad overviews. Multiple questions on individual conditions through all themes assimilated into comprehensive ideas centred on vertigo, dizziness and balance disorders. Tertiary prevention, in the form of vestibular rehabilitation, was incorporated into interventions under Clinical and Cost-Effectiveness. The group noted that a lot of work on service design and delivery had been done in other fields. Questions about service delivery were combined and, based on the general group discussion, a question was formulated about adapting work already done in this area from these other fields.
Infection, Inflammation and Allergy There is a need for acceptance and development of non-RCT research approaches including observational studies, for example on variation. Cultural change around equipoise and acceptance of randomisation, especially by surgeons, is also needed. Consideration needs to be given to equipoise in primary and secondary care and where patients are actually treated. Understanding variations in practice is important as it may give insight into barriers to care.	Multiple questions about otitis media, sore throat and rhinosinusitis were combined into overarching questions. Allergy and anosmia not included in Clinical and Cost-effectiveness questions: allergy research was being led by other groups, and anosmia was viewed as a symptom not a condition. Ideas were retained in developing pathways for allergy patients and in anosmia questions relating to assessing

Communication – improve patient understanding as well as patient clinician dialogue and consider how do we convey concepts of benefit v risk.
Education - the role of the pharmacist interesting in considering their role in modifying risk factors and reducing burden on primary care. Also education in primary care, especially in discouraging inappropriate prescribing of antibiotics.

impact, raising awareness and developing pathways from patient presentation.
Prevention – questions combined, and the role of dietary supplements added to the question as also potentially relevant.
Question of prevalence of rhinosinusitis excluded as existing evidence.
Multiple questions on understanding and diagnosing rhinosinusitis variants to determine care combined into defining populations for paths of care by endotypes and phenotypes allowing for targeted therapy.

Head, Neck and Airway
This was a process of distillation and it was felt this section was reasonably complete.
Improvement work happening in tracheostomy care but still a key area requiring research.
PROMs: COMET project is ongoing but will only come out with patient priorities, so more research to develop outcome measures for ENT, despite current research projects.
Could we improve research culture in Head and Neck? Although there is a good research culture in Head and Neck cancer there is still a need to improve capacity; a focus should be given to improving academic careers for SLT.

Exclusion of majority of head and neck cancer research ideas as already covered by the NCRI funded trials, including prevention, service delivery, rehabilitation, functional issues, and quality of life studies.
Functional outcome measures: outcome measures extended from head and neck cancer to include benign disease as they are also lacking here.
Pharyngeal pouches – question about stapling broadened to management as current NICE guidelines based on poor evidence.
Multiple questions about globus pharyngeus, OSA and snoring, voice problems combined into overarching questions. All noted to have little high-quality evidence, especially OSA and snoring.

Children and Young People
This has been distilled to generalisable themes, but it is recognised that there is a risk of it becoming too generic.
Poor evidence base noted with a need to fully evaluate research and evidence for current interventions and management. The concern with this is are outcome measures robust enough to make such evaluation meaningful?
Children with complex needs must be considered in research, evidence base lacking for their management.
Children grow, change and develop. Treatment and interventions need to change through childhood to reflect this. In developing evidence, consider the influence of the age of child to gain a clear understanding of relative benefit, and how this changes as a child develops.
Understanding of the natural history of common childhood illnesses felt to be under-addressed by research.

Questions through all themes combined into broad ideas focusing on common or high impact conditions and their management.
Clinical interventions such as hearing aids combined with non-clinical approaches, such as classroom interventions.
Development of clearly defined outcome measures lacking throughout paediatric ENT and linked to understanding the impact of these conditions on children, families and society. Questions around assessing this impact and developing outcome measures were thus extended to all common childhood ENT conditions.
Development of decision aids beyond those for glue ear, as concerns that even existing evidence not being used, thus widespread need to consider the process of delivering information to parents and clinicians and developing such aids.

An Ageing Population

Ageing population is not a single population; it is not homogenous, and it is changing.

It is rare that a person only gets to age with just hearing loss; consider instead the impact of multi-morbidity and polypharmacy.

Consider how people become patients, and the fundamental question of how older people are considered and how they think about themselves, especially around hearing loss – is this actually a normal part of ageing?

Research into balance conditions and service delivery for older people will require collaboration with other healthcare specialties and professionals, such as geriatricians, physiotherapists and occupational therapists.

A need to find and train people who want to research in this area was identified.

Multiple questions on hearing loss and balance in older adults were combined into broad questions about these conditions across the themes.

Questions about developing adult hearing screening programmes included here (see under Adult Hearing Loss and Tinnitus).

The loss of sensory function as a predictor for neurodegeneration was discussed by the Inflammation, Infection and Allergy group, but was felt to be relevant to this topic and so was included here.

Interventions for hearing loss strategies in an ageing population broadened beyond hearing aids, similar to discussion in Adult Hearing Loss and Tinnitus.

Table 22 Summary of Expert Forum key points and management of research ideas

	Inflammation, infection and allergy in adults	Head, neck and airway problems	Balance problems	Adult hearing loss and tinnitus	Children and young people	An ageing population	Total number of ideas
Understanding health, illness and disease	4	3	3	2	5	7	24
Prevention of ill health, maintenance of good health	1	1	0	2	0	1	5
Clinical and cost-effectiveness	4	9	3	8	8	4	36
Designing and delivering healthcare	5	2	3	2	2	4	18
Creating effective communication	3	1	0	2	1	0	7
Promoting knowledge and learning	2	1	2	1	1	1	8
Total number of ideas	19	17	11	17	17	17	98

Table 23 Number and distribution of short list of research ideas across the framework

5.3 The priorities

The participants in the Consensus Conference considered 98 research ideas and chose 21 priorities from these. The following sections give general discussion points from the groups and indicate the level of consensus as reported by the participants. These priorities were mapped back across the long list of ideas, stakeholder categories and framework, as shown in Tables 24 and 25.

5.3.1 Inflammation, Infection and Allergy priorities

Four priorities were chosen in this topic from 19 research ideas that were presented to the Consensus Conference. The group felt that these provided a linked continuum: clinical definitions of diseases were needed before thought could be given to treatments. This was summed up in the plenary session, with one of the group participants noting: "So, by choosing these four, we think we've got the start by doing the priority setting and definition of disease, the effective treatments and then important outcome measures and then our fourth thing would be looking at antibiotics as a slightly separate issue. " (Plenary session transcript, p 4: 130 – 133).

The group felt that there were two important ideas missing from this topic that appeared under other topics:

- the development of cell-based therapies for sensory ENT disorders, which they felt should be incorporated broadly to include sensory loss due to infection, inflammation or allergy;

- the patient representatives in the group emphasised the need for research into ways that patients could manage their own conditions or improve their care, again an idea that appeared in other topics, but which they felt was relevant here too (see Figure 22 for discussion notes captured by this group).

The area not prioritised by this group were specific questions about allergy. In the Expert Forum it was noted that colleagues in immunology and other allied medical specialties were already collaborating with ENT and primary care and had active research programmes in these areas. Participants in this group concurred, hence research ideas specifically around allergy were not highlighted as a priority.

There was a high degree of concordance with the different stakeholders in the small group discussion according to the participants. As one patient participant stated:

"I've been involved in a number of ranking type questionnaires, which have been filled in by clinicians and public or patients and it always surprises me how out of step those can be and the patients see things quite differently to clinicians at times, so it was quite a surprise in this one that we were very much in agreement on the top two items, surprised me considerably. The other items, there was a much broader spread of support for the questions, but that applied, I think, across the board. It wasn't an out of step thing

between the patients and clinicians." (Plenary session transcript, p 3: 98 –

104).

Another participant agreed, stating that they "Really want to say that we had a

really, really good discussion" with "harmony between the medics and the

patients" (Plenary session transcript, p 4: 149 – 150).

5.3.2 Head, Neck and Airway priorities

This group discussed 17 research ideas across all the themes and chose four

priorities to present to the plenary session. In this group, the participants

presented their top choices to the group before further discussing them. The

group facilitator noted that there was clear concordance in the group; the four

priorities chosen had been independently highly rated by all the participants

(HNA Group transcript, p 8: 353 – 370).

Figure 22 Photograph of notes made by participants during Consensus Conference, small group work Inflammation, Infection and Allergy

The final priorities agreed by this group all appeared to be from the same theme of Clinical and Cost-effectiveness. The participants explained to the full group in the plenary session that this did not reflect the true nature of the discussion:

"…I don't think that quite summarises the depth of our discussion actually. I think what was quite interesting, now forgive me, if anyone in my group thinks I'm misrepresenting what happened in the group, but it seemed like when we first went round, we had some fairly clear ideas and actually the more we unpicked things, the more we moved around. Actually we had, although we ended up with all four (unclear 0:04:32) spots, there was a lot of discussion towards the end about patient pathways, about communication and things like that, but actually at the end it was then felt that we could encompass all of those things, although they were still important, but actually we could cover all of those by answering these questions." (Plenary discussion transcript, p 2: 50 – 58).

5.3.3 Balance Problems priorities

This group discussed 11 research ideas and chose three priorities. In their feedback to the whole conference, one of the participants in this group commented on the consensus they had reached. They noted that different stakeholders had different perspectives, but felt that these were acknowledged and incorporated into the final decisions: "…the clinicians were worried about the diagnosis and the patients were more worried about the

patients' experience, although I think it's probably best to say that both groups appreciated the value of both parts." (Plenary Discussion transcript, p 6: 271 – 273).

5.3.4 Adult Hearing Loss and Tinnitus priorities

This group discussed 17 research ideas across all the themes. They chose four priorities. One of the patient participants considered the degree of consensus and the concordance in this group. They noted that there had been considerable debate, but fundamentally felt that there was accord, stating "the clinicians and myself were rather in agreement which is something that I'm taking away from this which is very positive." (Plenary Discussion transcript, p 7: 333 – 348).

5.3.5 Ageing Population priorities

This group discussed 17 research ideas across the framework and prioritised three. They noted that they hadn't had the same degree of agreement as the other groups at the beginning of their session when presenting their initial ideas about priorities:

"So we were perhaps slightly apart from the other groups because when we put all priorities down, I think we covered almost every question that we had [laughter] in the list. So I think the struggle we had within our group, was really trying to decide what was the most important, because it all seemed so

important and do correct me if you disagree." (Plenary session transcript, p 5, 182 – 185).

Part of the difficulty for reaching consensus with this group was challenging different perceptions as to what was meant by "an ageing population":

"I think the only thing we should reflect on is that we had quite a discussion about what was meant by an ageing population, so did we mean the effects on services of having a population in general which is getting older but who may be a very fit population with their biological age or their physiological age or were we looking at the effects of having a larger very elderly population with particular needs, so that's just a, kind of, we think it's a bit of both." (Plenary session transcript, p 5, 213 – 218).

The group did not think this population should be limited to those with increasing multimorbidity and polypharmacy. Instead it was recognised that this was both a heterogenous and a changing population (a point that also raised in the Expert Forum):

"I think, conversely, if you think of an ageing fit population, which is perhaps not the population we're seeing 20 years ago, you see I would say average 75-year-old these days is pretty fit and actually some of it might not be complexity around multi morbidity, but complexity about expectations and quality of life because your 75-year-old still wants to be playing basketball and actually the dizziness is having an extremely large impact on quality of life

compared to perhaps a 75-year-old 20 years ago who may have been less reliant on mobility, will have more boundaried horizons. So, I think there's that two branches, many more, but at least branches of complexity with an older population and what priorities are for what health looks like." (AAP Group transcript, p 5 – 6: 180 – 188).

Acknowledging this complexity informed the approach that this group took towards deciding their priorities. They felt that a strategic approach towards developing an agenda, as opposed to simple priorities, was needed and consequently they chose broad but linked ideas that had both short and long term impacts.

5.3.6 Children and Young People priorities

This group discussed 17 research ideas across the themes and chose three priorities to present to the plenary session. The diagnosis and management of OSA in children was the only condition-specific idea that was prioritised by the group. They felt that other conditions were equally important but noted that there were separate topics for Hearing Loss and Tinnitus and Balance Problems, albeit for the adult population. Participants in the group felt that this narrowed the priority options for children more than for adults: "And we've only got these, so it seems like we're having a raw deal that we have to pick two for the whole of the childhood population where everybody else gets that six groups out there, four or five groups out there." (CYP Group transcript, p 3: 183 – 185).

The group suggested that the priorities in these other areas should be applicable to the paediatric population. This was expressed in their feedback to the plenary session.

5.3.7 Mapping Consensus Conference priorities to the stakeholders and framework

To determine if the views and ideas of a spread of stakeholders were represented, or if one stakeholder category had taken precedence through the process, the priorities agreed at the Consensus Conference were mapped back to the original data from the survey and focus groups. Table 24 shows the spread of these priorities across the survey, focus groups and stakeholder categories.

In order to assess for bias the short list of ideas developed from the Expert Forum was re-entered into the framework (see Table 23, page 138). The rationale for this was that the framework had been rigorously developed from the whole of the data; if themes were neglected it would indicate that ideas weighty enough in the data to form these themes had been excluded by the narrower group of stakeholders prioritising them. This was also done after the Consensus Conference. The spread of the priorities following that is shown in Table 25.

Priority	No. survey responses	Focus Group number	Stakeholder categories
Define the different variations of disease that make up rhinosinusitis (endotypes and phenotypes) and develop ways of classifying them based on signs, symptoms and biochemical markers.	31	2, 3	Patient/carer Healthcare professional (4 types) Allied sector Industry
What are the most clinical and cost effective treatments for chronic rhinosinusitis?	42	4	Patient/carer Healthcare professional (5 types) Allied sector Service commissioner
How do we improve the implementation of guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden and its impact on antibiotic resistance in adults and children?	6	4	Patient/carer Healthcare professional (2 types) Allied sector
What outcome measures and instruments should we use in primary and secondary care for chronic ENT infections and inflammatory conditions?	5 (specific to IIA) 12 (general outcome measures)	2, 5	Healthcare professional (1 type) Allied sector Service commissioner Patient/public Healthcare professional (3 types) Allied sector/Industry
What is the optimal management for adults with OSA?	8	4	Patient Healthcare professional (2 types) Allied sector
What outcome measures and instruments should we use for research into head, neck and airway disease, both benign and malignant?	9 (specific to HNA) 12 (general outcome measures)	2	Patient Healthcare professional (4 types) Patient/public Healthcare professional (3 types) Allied sector/Industry
What is the role for improved diagnostics in head, neck and airway conditions?	13	2	Patient Healthcare professional (4 types)
What is the optimal management for adults with voice problems?	11	2, 3	Patient Healthcare professional (3 types) Allied sector/Industry
What is the optimum pathway for people with balance problems? What can be dealt with in primary care and the community, and what are the indications for referral? How should specialist clinics be set up?	53	1, 2, 3, 4, 5	Patient/carer Healthcare professional (7 types) Allied sector Service commissioner/manager

How can patients be empowered to self-manage their balance problems and how does this impact on their outcomes?	8	3, 4, 5	Patient Healthcare professional (2 types) Allied sector Service commissioner/manager
Develop clear diagnostic criteria for different balance disorders so that patients can be informed about prognosis and best treatment.	28	1, 3, 4	Patient Healthcare professional (7 types) Allied sector
What are the best management strategies for tinnitus?	31	4	Patient Healthcare professional (7 types) Allied sector Service commissioner Research funder
Which hearing aid strategies work best for adults with hearing loss?	55	1, 2, 5	Patient/carer Healthcare professional (8 types) Industry Service commissioner Allied sector Research funder
What is the burden and impact of hearing loss on an individual, family and population level?	44	1, 3	Patient/carer Healthcare professional (3 types) Allied sector Service commissioner Research funder
What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?	35	2	Patient/carer Healthcare professional (5 types) Allied sector/Industry
What are effective interventions for hearing loss in older people?	19	1, 5	Patient/public Healthcare professional (2 types) Service commissioner/manager Research funder
Is it beneficial to introduce an adult hearing screening programme and how do we best develop it?	10	2	Patient Healthcare professional (4 types) Service manager Research funder
What is the optimum design for "balance pathways" in older people with balance problems? How and where can older people with balance problems and with multiple medical conditions and decreased mobility	24	1,3,4,5	Patient/carer Healthcare professional (5 types) Allied sector Service commissioner/manager

best be managed? How can self-management strategies be built into these pathways and what is their impact on patient outcome?			
What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?	32	1, 2, 3	Patient/carer Healthcare professional (9 types) Allied sector/Industry Service commissioner/manager Research funder
How do we best diagnose and manage obstructive sleep apnoea (OSA) in children?	9	N/A	Patient Healthcare professional (2 types) Allied sector
How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professionals in different settings, and what is the impact of this on patient outcome?	13	1, 4	Patient Healthcare professional (6 types) Allied sector Service commissioner/manager

Table 24 Mapping priorities to survey, focus groups and stakeholder categories

	Inflammation, infection and allergy in adults	Head, neck and airway problems	Balance problems	Adult hearing loss and tinnitus	Children and young people	An ageing population
Understanding health, illness and disease	1			1	(1)*	
Prevention of ill health, maintenance of good health	(1)*					1
Clinical and cost-effectiveness	1	4	1	3	2	1
Designing and delivering healthcare	1		1			1
Creating effective communication			(1)**			
Promoting knowledge and learning			1		1	

Table 25 Number and spread of Consensus Conference priorities across the framework

*Idea taken from theme and incorporated by the Consensus Conference group into another priority

**Area of disagreement: analysis placed priority here, Consensus Conference placed it into Promoting Knowledge and Learning

5.4 Summary of priorities and reasons for their prioritisation

This chapter has shown the development of the long list of ideas and how the short list evolved from that. It has also demonstrated how the research priorities were determined by stakeholder discussion in the Consensus Conference with the reasons for prioritising them. Combined with the previous chapter this is the full presentation of the findings for the RPSE. Table 26 below summarises each of the individual priorities, whether they were altered from the list developed from the Expert Forum, specific reasons for prioritising them and other key discussion points.

The next chapter discusses the strengths and limitations of the exercise, compares these results with the previous topic-specific exercises, and reflects on the challenges of setting research priorities.

Priority	Origin and alteration	Reason for prioritising	Discussion notes and illustrative quotes
Inflammation, Infection and Allergy			
Define the different variations of disease that make up rhinosinusitis ("endotypes" and "phenotypes") and develop ways of classifying them based on signs, symptoms and biochemical markers.	Taken directly from list given to participants; no alteration.	Lack of basic understanding of disease and poor dissemination of pre-existing evidence results in late diagnosis exacerbating conditions. A robust research agenda should balance basic science and clinical research.	This priority highlights the need for 'personalised medicine,' which was felt to be important in a research agenda.
What are the most clinical and cost-effective treatments for chronic rhinosinusitis?	Idea modified; detailed list of treatments cut from final priority as it was long but not exhaustive and "because we feel that if you have that broad umbrella then you can define different treatments as you go along with the research." (Plenary session transcript, p 3 – 4: 118 – 120).	These conditions are common in primary and secondary care. There is a large variation in management of these conditions and inconsistent advice for patients.	What was highlighted in the group discussion was that although this broad approach was taken, the group felt that it was important that specific topics did not get lost. Ideas around prevention and treatment of anosmia were specifically requested to be incorporated into this priority. "One key ENT problem that we felt was important to highlight within that, was anosmia as a clinical problem which can be linked to other diseases and which came through very strongly from the patient representatives in our group as something that is felt to not get much air time from us clinicians but also hugely be important in terms of what they'd like." (Plenary session transcript, p 4: 133 – 137).
How do we improve the implementation of guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden and its impact on antibiotic resistance in adults and children?	Ideas combined: adapted from ideas around antibiotic use in specific infections.	Antibiotic misuse and resistance recognised as a key issue for the NHS and wider society.	This was the only issue that the group felt was important enough to retain on its own instead of incorporating into a wider question about the implementation of guidelines. The group was challenged on this by the facilitator but felt that the issue of antibiotic misuse was too important an issue in its own right.
What outcome measures and instruments should we use in primary and secondary care for chronic	Ideas combined: adapted from ideas about outcome	Research aims and outcomes need to be agreed to develop high value research.	There is a need to capture important outcomes to patients especially those with "hidden" sensory loss, such as the impact on the senses of smell and taste.

ENT infections and inflammatory conditions?	measures for specific conditions.		
Head, Neck and Airway			
What is the optimal management for adults with OSA?	Taken directly from list given to participants; no alteration.	Large clinical burden of disease, and the potential to develop appropriate pathways that avoided inappropriate referral to secondary care. Significant impact on health, quality of life and safety of individuals.	"In terms of employment of patient, I think that's been a big issue because sleep apnoea is one of the common causes of daytime sleepiness and road traffic accidents and it's the biggest problem in long distance lorry drivers." (HNA Group transcript p 7: 297 – 299).
What outcome measures and instruments should we use for research into head, neck and airway disease, both benign and malignant?	Idea modified: expanded to include benign and malignant disease.	Agreed outcome measures will improve the value of research investment and ensure that the results are relevant to patients.	"I think a lot of research is hampered full stop but particularly in head and neck and ENT, with not having core outcome measures agreed beforehand everyone's measuring different things, therefore generalising results, so everyone's planning their own outcomes and measuring different things and actually you end up not really being able to put all that data together." (HNA Group transcript, p 4: 145 – 150). Feedback to Plenary session: "I think if we were able to define better, what is important for patients in particular, that would be very useful." (Plenary discussion transcript, p 3: 64 – 70).
What is the role for improved diagnostics in head, neck and airway conditions?	Idea modified: originally related to the role of improved diagnostic technology in head and neck cancer. The group expanded it, as it was relevant to other ideas relating to benign conditions.	Improving diagnostics will have an impact on patient care, outcomes and quality of life.	"There was strong feeling that voice problems and voice treatment and diagnostics were very important and unrepresented in research in general, but actually and so initially we had quite a lot of voice questions in here, but it was felt that not only from the outcomes, but also from improved diagnostics, that would improve a lot of the problems that patients have with their voice problems." (Plenary discussion transcript, p 3: 71 - 77). Not all group participants felt that research into using improved diagnostics was an appropriate use of resource however. Some of the clinicians were concerned that the real-world impact of such research would be limited by the realities of what Trusts were prepared to fund. After

			discussion it was felt that expanding the question to incorporate benign disease represented the concerns expressed by many of the stakeholders around the management of these problems. Thus the group reached consensus on keeping this as a priority.
What is the optimal management for adults with voice problems?	Taken directly from list given to participants; no alteration.	Voice problems are a substantial area of unmet need for patients.	"People with voice problems tend to shy away from social occasions, they're uncommunicative, that also can lead to further health problems, depression, isolation, all those other aspects. So that's something I feel quite strongly about. Personally I have a couple of friends that have problems with their voice; they've never sought help for it, even though we've tried to tell them to because they don't really know where to go. They don't really know what facilities are available to them and they just think it's the way it is; they drink too much coffee, too much wine, whatever." (HNA Group transcript, p 5 - 6: 220 - 227). There was discussion within the group as to whether, by expanding the idea about improved diagnostics, it was still necessary to also include a question about the management of voice problems. However this was an issue that the patient participants strongly advocated and thus it was agreed it was a priority.
Balance Problems			
What is the optimum pathway for people with balance problems? What can be dealt with in primary care and the community, and what are the indications for referral? How should specialist clinics be set up?	Idea modified: the sub-questions under this idea were based on ideas incorporated from two other themes: Clinical and Cost-effectiveness and Promoting Knowledge and Learning.	Management and care need to be centred around patients with a focus on the problems of the patient, not on clinical specialties or a single body system. Lack of access to services and delay in diagnosis causes distress to patients and lengthens their experience of illness.	All group participants rated the idea about balance pathways highly. "It's how we manage to provide as good as cost effective service as we possibly can and maybe good research needs to be put into that, to find if it is cost effective, because if you do find out that well, if we run this service, then it saves appointment x, y and z, it also gets this person back to work, it stops this social care bill because we don't need this, four times a day care, twice a day care, whatever it is, then that gives you the business case to push forward for these things." (BP Group transcript p 13: 640 – 646).

How can patients be empowered to self-manage their balance problems and how does this impact on their outcomes?	Idea developed based on existing ideas in their own and in other topics. Extrapolated from a Promoting Knowledge and Learning theme idea that asked: "What is the impact on patient safety of raising public awareness of balance conditions?" Addressing lack of awareness would be a key step in self-management and have an impact on outcomes.	Improving the ability for patients to self-manage, including online and peer support, since balance problems are often chronic.	When feeding back their priorities to the wider group in the plenary session, they noted that: "We spent quite a bit of time talking about patient experience of balance disorders and the resource that there is in that patient experience and how patients can support each other and the very distressing experience of acute balance disorder or long term management of chronic conditions and how to best use that." (Plenary Discussion transcript, p 6: 234 – 237). By grounding this idea in the other ideas that had been proposed from the data, the group agreed that this was a suitable priority that reflected both the data and their own experiences.
Develop clear diagnostic criteria for different balance disorders so that patients can be informed about prognosis and best treatment.	Taken directly from list given to participants; no alteration.	Diagnosis is important both for patient benefit and developing future research of existing and novel therapies. Balance conditions are difficult to diagnose, and more research is needed to understand individual causes and patterns of balance problems to inform diagnostic criteria.	Favoured by healthcare professionals. They recognised that although there were pre-existing diagnostic criteria, more research was needed as: "…even within a group like Vestibular Migraine or Meniere's Disease, the patients are very varied, the problems that they have and the symptoms that they have, the things they are bothered by, and I suspect that there are different sub-groups in those different diagnostic criteria that would benefit from different kinds of treatment approaches. So sorting out the causes and the course, and the diagnoses is all part of the same problem." (BP Group transcript, p 8: 353 – 357).
Adult Hearing Loss and Tinnitus			
What are the best management strategies for tinnitus?	Taken directly from list given to participants; no alteration.	Tinnitus is a common, often hidden condition with underestimated impact; research has been under-resourced and there is a lack of evidence for best management.	This had clear consensus from the group: "It's a fair burden on our ENT Service and Audiology Service and it's often quite frustrating. I think that everyone manages it slightly differently and we just don't seem to have the right answers for it. I think with such a common problem, that if we can understand it a little bit more and know what the

		Rapid gains from research possible.	evidence behind that was, I think that would help the NHS all in all." (AHLT Group transcript, p 8; 387 - 391).
Which hearing aid strategies work best for adults with hearing loss?	Idea modified: different ideas combined into a broader priority.	Hearing loss is a common, growing problem affecting large numbers of people. The uptake of hearing aids is low, and many people are unhappy with the benefit they offer; research into new devices and technologies would help us better meet population needs and could provide rapid benefits.	This priority took negotiation in the group, as there were other questions relating to auditory implant devices and to hearing aids for specific groups of the hearing impaired. The group agreed that the broad question about hearing aid strategies could incorporate these other questions, stating "that devices and technology overlap and for argument that would be one area." (AHLT Group transcript, p 16: 829 – 830).
What is the burden and impact of hearing loss on an individual, family and population level?	Taken directly from list given to participants; no alteration.	The better the understanding of burden and impact of hearing loss, the more value research and services based on this understanding will have, as it will be informed by patient experience and clinical need.	There was initial disagreement: "Can I just offer a word of caution. Looking at other sections they've all got something similar and it reminds me of what we have done with inequalities in health for the last 40 years. We've spent millions on research describing inequalities and absolutely sod all doing anything about it. If this is about investing in research, it would be very easy to spend money describing how awful it is as a result, but it doesn't mean we're going to achieve a lot." (AHLT Group transcript, p 16: 855 – 860). After discussion the participant noted that "focusing on the burden on society can work occasionally" if it influenced those in political power to provide more resources for research by raising the profile of the issue (AHLT Group transcript, p 19: 1025 – 1038). Another participant had asked colleagues for their opinions and was surprised they had chosen this as a priority as he had not considered it but was swayed by their views. After this discussion it was presented as a priority to the plenary session.
What are the effects and costs of novel medical and biological treatments (such	Idea modified: costs of treatment included in the scope.	Long-term aspirational priorities, such as research into novel hearing treatments should	One participant felt that the gains from research were too far off to be part of a research agenda: "…the reason why I didn't go down the stem cell therapy is that at the moment there is some fantastic work especially going

as cell-based therapies) for sensorineural hearing loss?		balance shorter-term research gains.	around in the Manchester area but that is more relevant for regenerating nerves. But regenerating cochleas? For nerves we are about 10 maybe 15 years away, regeneration for cochlear hair cells is not even on the horizon." (AHLT Group transcript, p 11: 571 – 576). Other stakeholders felt that "It would be quite nice to have some real science" (AHLT Group transcript, p 21: 1110), and one of the patient participants positively described this sort of research as aspirational. Ultimately the group consensus was that "We have some quick fixes. But looking at the big picture is probably a good idea" (AHLT Group transcript, p 21: 1136).
Ageing Population			
What are effective interventions for hearing loss in older people?	Taken directly from list given to participants; no alteration.	Effective interventions for age-related hearing loss would impact on a large group of people, improving individual quality of life and population health.	"As you reach my age you will find that you have a significant hearing loss even though you are living well and have no medication and have no health issues, but your hearing will be less than it is at your age, and that whole issue of an aging population as opposed to an old or elderly or geriatric, whatever that...and this is a question obviously I have is that are we thinking in this subgroup, are we thinking about that aging population, and I'm going to put myself in that aging population; are we thinking about the elderly, and I think that's a very gradual continuum, and physiological age is one aspect of it, but there's that huge population of people who are losing their hearing, who have nothing else going on with them." (AAP Group transcript p 6: 189 – 197).
Is it beneficial to introduce an adult hearing screening programme and how do we best develop it?	Taken directly from list given to participants; no alteration.	These interventions for age-related hearing loss combined with the public health intervention of an adult hearing screening programme could transform expectations of quality of life and have a significant	"I suppose it comes back to what you were saying: they're all tied together. If we are assessing a research agenda, maybe if we think that we should be looking at more on a national strategic basis of having a screening, hearing screening programme, what sort of research questions that have to be answered in order to make that case, so perhaps that's a good way of looking at it, so then we could say, well, actually should you be saying to

		impact on vulnerable populations.	researchers, well, looking more at how can we improve hearing loss, how can we restore hearing, and setting that agenda for researchers in order that we could get benefit from early identification, do you see what I mean? Maybe that's a way to get around the complexity. I don't know." (AAP Group transcript, p 14: 552 – 560).
What is the optimum design for "balance pathways" in older people with balance problems? How and where can older people with balance problems and with multiple medical conditions and decreased mobility best be managed? How can self-management strategies be built into these pathways and what is their impact on patient outcome?	Idea modified: different ideas combined to give a broader priority.	Balance pathways in older people can have an impact on quality of life and on the health burden of falls, and need to consider the potential for multiple medical problems.	Self-management was felt to be important and applicable to people with many conditions, including balance problems, with one participant noting "I think self-help has a big role in helping people through any, not just ageing or any ailment, but any dizzy person to improve their balance" (AAP Group transcript, p 14: 584 – 591). It was noted that self-management had had an impact on other areas of healthcare, and that research in this area might have wider impact.
Children and Young People			
What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?	Idea modified: divided into impact for the two different populations as most of the studies exclude children with complex resulting in little evidence for that population. Many other ideas in this topic acknowledged the two groups so that was adopted here (CYP Group transcript, p 19:874 – 877)	To improve the value of future research the impact of conditions on children and families needs to be better understood. Research outcomes need to be agreed upon by researchers, clinicians and parents so that research is patient-centred, and results can be compared across studies.	"Most of the stuff we do in children's ENT is not about saving lives. It's about improving the quality of people's lives and it's about all of this stuff and we don't measure that very well. The outcome measures that we use in our daily practice and in research and all the rest of it is numbers on a chart. It's a hearing test, so you have a hearing test and it's better or it's not better and here's some numbers but what that absolutely doesn't capture is people's day to day experience, the things they struggle with: socialising, education, all the rest of it. And we don't capture those things well and we don't measure them, and you can't then do any meaningful research that shows whether what you're doing is useful or not unless you capture the things that are important to you, the things

			that they're actually coming to see you about." (CYP Group transcript, p 7 – 8: 317 – 327).
How do we best diagnose and manage obstructive sleep apnoea (OSA) in children?	Taken directly from list given to participants; no alteration.	OSA is of great concern to parents and can have a significant impact on children and their development and school achievement; there is uncertainty and variation in how it is best managed.	This was only condition-specific idea prioritised by the group and was proposed by healthcare professionals. It was singled out due to the level of concern about this condition from parents in terms of impact on development and school performance, which were not represented in the adult topic.
How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professionals in different settings, and what is the impact of this on patient outcome?	Idea modified: the top two ideas chosen by all participants were combined.	ENT, Hearing and Balance conditions are very common in children. Knowledge of ENT, Hearing and Balance conditions is variable in different health care settings, despite the large burden of disease of these conditions.	Three patient/patient carer participants had prioritised this. They felt that outside of specialists this knowledge was lacking and that it had an impact on the timeliness of treatment and consequently on outcomes. Although none of the healthcare professionals had originally selected this as a priority, through discussion with the other stakeholders and sharing their own experiences of insufficient training in ENT, they agreed that research in this area was a priority. They noted that it had the added benefit of being "potentially a quick win from a research point of view" (CYP Group transcript, p 14: 613).

Table 26 The priorities with reasons for prioritisation and discussion notes

Chapter 6: Discussion and Conclusions

6.1 Introduction

This study used qualitative methodology in a multi-step process to address the overall research question:

What are the research priorities for ENT, Hearing and Balance care in the UK according to the views and voices of its stakeholders?

In this final chapter, the findings of the previous chapters will be summarised, and their empirical and methodological contributions will be considered and discussed in the context of the literature. Areas for further research, and the impact of this thesis on research activity and implications for policy will then be discussed. Following this, there will be reflection on the study design and its limitations, and the chapter will conclude with critical reflections, including a reflexive examination of the role and impact of the author of this thesis.

6.2 Summary of main findings

The scoping review in Chapter 2 found that according to the literature there is no "right" way to develop a research priority setting exercise. Rather than using a specified method, for the exercise to be robust the quality of the process developed needs to be considered. Transparency, inclusion and credibility were identified as indicators of quality to be considered when designing a process. Many different methodologies and methods used in

RPSEs were identified. Qualitative methodology using multiple methods was determined to be the approach that satisfied the identified quality criteria for a multi-stakeholder exercise that aimed to capture their views and voices. Previous RPSEs in ENT, Hearing and Balance care were also identified in the literature, but none of them addressed the full scope of the question asked in this thesis.

The third chapter of this thesis detailed the methodology, process and methods used to design the RPSE that produced the findings of Chapters 4 and 5. Chapter 4 determined the stakeholders for UK ENT, Hearing and Balance care. Six stakeholder categories were identified based on the literature, expert opinion and stakeholder consultation: patients and public; healthcare professionals; managers and service commissioners; allied sectors; industry; and research funding bodies.

Chapters 4 and 5 both addressed the primary research question of this thesis, as given above. Chapter 4 described the framework for UK ENT, Hearing and Balance care research priorities inductively developed from stakeholder responses. This was a two-dimensional matrix of interpretive themes and descriptive clinically-oriented topics. Six broad research themes were identified, extending beyond the clinical effectiveness research ideas that had been initially considered for this exercise. Analysis of the survey responses and focus group discussions revealed that they also crossed six topics, based on ENT, Hearing and Balance problems and the different age groups affected by them. These formed the second dimension of the framework.

Chapter 5 provided the 21 research priorities developed and chosen from an initial 1697 research ideas; mapping these back to stakeholders and the framework showed a range of stakeholder categories contributed to each priority, though the priorities tended to cluster in the Clinical and Cost-effectiveness theme (see discussion below). Finally, this chapter also gave detailed descriptions of the reasons for prioritising these, according to UK ENT, Hearing and Balance care stakeholders. These considered impacts on multiple areas (individual patient, society, healthcare systems and knowledge basis) and are discussed in more detail below.

These main findings make empirical and methodological contributions to the literature on research priority setting. To assess these contributions, the findings will now be summarised and considered in the context of the literature in the next section.

6.3. The empirical and methodological contributions of this thesis – priorities, purism and pragmatism

Based on the findings outlined above this study makes three empirical contributions. Firstly, it defines who the stakeholders are for UK ENT, Hearing and Balance care, according to the stakeholders themselves. Previous literature for the UK exercises had used pre-defined categories, which had not allowed for greater exploration of those who could have a vested interest in the prioritisation process (the definition of a stakeholder according to

Campbell 2010). Rather, concerns about historical imbalance in influence and bias in process had dictated the categories. This study has widened the consideration of who the stakeholders are in a UK context.

Secondly, this thesis has presented the only study to date of ENT, Hearing and Balance care research priorities done across the whole field and a wide range of stakeholders in the UK. In doing this it has extended the concept of what types of research priorities should be developed, based on the views of the stakeholders. This was done by inductively developing the framework for the priorities. The previous exercises (Hall *et al* 2013; JLA PSP 2011; van Benthem *et al* 2013) had predetermined the areas of inquiry and thus did not fully explore where stakeholders' priorities could lie outside of traditional treatment uncertainties. This thesis demonstrates the range of research stakeholders want to consider when setting research priorities. The 21 priorities developed have contributed to the existing knowledge, which has either been confined to discrete clinical topics (Hall *et al* 2013; JLA PSP 2011), been conducted in another country (van Benthem *et al* 2013), or both (Patel *et al* 2013; see note on updated literature review below).

The third and final empirical contribution is to the understanding of why the stakeholders have chosen the priorities, both for ENT, Hearing and Balance care as well as for other topics. The RPSEs for the former have either not given these reasons (Hall *et al* 2013; JLA PSP 2011), or the criteria have been pre-set (van Benthem *et al* 2013; Patel *et al* 2013). This study also adds to reasons identified in the wider literature.

This thesis makes one methodological contribution to the literature. Through the findings in the second Chapter, the three parameters of quality for research priority setting processes were identified: transparency, inclusion and credibility. The exercise designed for this thesis used purely qualitative methodology with framework analysis in order to fulfil those criteria, an approach not previously described in the literature.

These contributions will now be considered in more detail and in the context of the literature on research priority setting. The research priority setting exercise that formed the basis of this thesis was planned in 2013, started in 2014, and concluded in December 2015. Research priority setting was a dynamically evolving field through this time. In order to discuss the thesis in the context of the literature, following the exercise the review described in Chapter 2 was updated with an end date of 31 March 2016. This extended time period allowed for this RPSE to be considered with contemporaneous developments in understanding how to set research priorities, and what those priorities were for ENT, Hearing and Balance care. The methods and findings of that update are summarised in Appendix 2, page 224.

6.3.1 Empirical contributions: stakeholders

The previous UK research priority setting exercises in ENT, Hearing and Balance care had engaged pre-set categories of stakeholders: patients, carers and clinicians. In the updated literature review, a further American exercise was identified. This was carried out by the American Agency for

Healthcare Quality and Research for Obstructive Sleep Apnoea (Patel *et al* 2013), which was again topic specific. This exercise did consult a broader range of stakeholders based on categories, similar to the exercise in this thesis.

Six categories were described in Patel *et al* (2013): patients and the public; providers; purchasers of health care; payers; policymakers; and principal investigators. Those categories were pre-determined and very similar to those described by Nasser *et al* (2013), so they were not derived in consultation with stakeholders. What is missing from that taxonomy are the stakeholders defined by Allied Sectors, Industry and Research Funding Bodies. Specifically, in this study, Allied Sectors embraced potential stakeholders outside of traditional healthcare professionals who could be considered to have an interest in the outcome. Professionals, such as Teachers of the Deaf, may have considerable input into the health outcomes of patients. Gaining this insight from the stakeholders in this exercise challenged these preset ideas of who could contribute to developing priorities and thus prevented exclusion of relevant voices.

6.3.2 Empirical contributions: the priorities

As was seen in the literature in Chapter 2, there have been previous research priority setting exercises in ENT, Hearing and Balance care both from the UK and abroad. One further UK exercise and an American exercise were identified in the updated review (Henshaw *et al* 2015; Patel *et al* 2013). The UK exercise again used Priority Setting Methodology and was topic-specific

(mild to moderate hearing loss). The American exercise extended to a broad range of stakeholders, as described in section 6.3.1 above, but was also topic specific.

It is interesting to compare the outputs of other exercises with those from the present exercise. Table 27 shows the priorities of the previous UK Balance Priority Setting Exercise (JLA PSP 2011) exercise with those from similar topic areas in this exercise. The Balance PSP developed a list of the top ten research priorities based on the views of patients and clinicians. As can be seen from the table, the PSP priorities were a mix of broader questions relating to balance in general, and more specific ones with a focus on conditions (vestibular migraine; Meniere's disease) and interventions (home-based exercises; stress management techniques).

There were similar findings in the previous Tinnitus PSP (Hall *et al* 2013; see Table 31, Appendix 4, page 237). The top priority in this PSP was: "What management strategies are more effective than a usual model of audiological care in improving outcomes for people with tinnitus?" This was similar to the one tinnitus priority that was developed in this exercise ("What are the best management strategies for tinnitus?"), which was also a broadly phrased idea. Seven of the other top ten PSP priorities also related to the management of tinnitus, but for specific populations (children; people with profound hearing loss), symptoms (tinnitus-related insomnia) or interventions (CBT; complementary therapies; hearing amplification; medications).

This combination of wider ideas with more specific questions is similar to what was seen in the exercise developed in this thesis, as was discussed in the previous section. This reinforces the challenge found in this exercise of developing shared priorities from different types of stakeholders whilst retaining sufficient detail to inform potential research questions, which is discussed further under methodological contributions (section 6.3.4).

What can also be seen is that some of the priorities from the Balance PSP address research ideas extending beyond treatment uncertainties or prevention, testing and rehabilitation, the defined scope of PSPs (Cowan and Oliver 2013). These priorities considered educational strategies and the potential for service provision outside of secondary care. They would potentially fit under different themes in the framework developed in this thesis (Promoting knowledge and learning; Designing and delivering healthcare). This indicates that stakeholders' priorities may lie outside that traditional scope, and confining them to those preset areas may not give a true indication of research that they consider important.

James Lind Alliance Balance PSP 2011	Priorities from GENERATE
What is the optimal process for GP education and training for improved diagnosis and management of balance disorders?	Develop clear diagnostic criteria for different balance disorders so that patients can be informed about prognosis and best treatment.
What is the best way of training health professionals in the management of balance disorders?	
What is the most effective treatment for vestibular migraine?	What is the optimum pathway for people with balance problems? - What can be dealt with in primary care and the community, and what are the indications for referral?
What are the best interventions to improve balance/minimise symptoms in daily activities such as supermarkets, escalators etc.?	- How should specialist clinics be set up?
Is any specific surgical intervention effective in Ménière's disease and what procedure is best?	What is the optimum design for "balance pathways" in older people with balance problems?
Are there any effective interventions for the ear pressure symptoms in Ménière's disease?	-How and where can older people with balance problems and with multiple medical conditions and decreased mobility best be managed?
What is the optimum pharmacological strategy for the management of patients with Ménière's disease? In particular, what are the effects of betahistine (including long term effects)?	-How can self-management strategies be built into these pathways and what is their impact on patient outcomes?
Is it helpful in preventing the severity, frequency and progression of attacks of Ménière's disease to adopt a specific diet, or restrict salt, caffeine or fluid intake?	How can patients be empowered to self-manage their balance problems and how does this impact on their outcomes?
Are the home-based exercises given to patients with balance disorders effective?	
Are stress management techniques helpful in patients with balance disorders?	

Table 27 Comparison of balance priorities

The final form in which the research priorities were presented in the Dutch research priority setting exercise (van Benthem *et al* 2013) was similar to that developed in this thesis. They were presented in a two-dimensional matrix of ENT sub-specialty areas and three research themes: Primary research questions, Guidelines and Other. This exercise covered all of ENT rather than a single topic. This demonstrates the utility in using a framework when considering priorities across a therapeutic field, where large volumes of data are likely to be generated.

The actual form of the final priorities was however very different compared to the exercise in this thesis and the other previous UK exercises. The priorities were very specific, particularly those in the first theme of research questions. The Dutch exercise prioritised questions such as:

"What are the factors that determine the effectiveness of speech understanding in background noise with:

- bilateral hearing aids,

- hearing aid and cochlear implant,

- or bilateral cochlear implants?" (See Table 31, Appendix 4, page 237).

This can be compared to an equivalent question from this exercise: "Which hearing aid strategies work best for adults with hearing loss?" Clearly these are thematically similar, but the former is more technical. This would appear to reflect the stakeholders from that exercise. These consisted of ENT

physicians and Patient Organisations. With the latter there was no indication that the Dutch exercise attempted to recruit "unorganised" patients. Thus, the technical language may have been suitable for their stakeholders, whereas it would likely have proved a barrier to engaged participation in the exercise in this thesis.

The American exercise (Patel *et al* 2013) has been discussed under the stakeholder section above. In that exercise they identified and prioritised topics for the diagnosis and treatment of obstructive sleep apnoea (OSA). These were broader topic areas, such as "cost-effectiveness of management strategies," rather than detailed research questions. Again, this may reflect the broad engagement across stakeholder categories in that exercise, compared to the detailed approach in the Dutch exercise.

6.3.3 Empirical contributions: reasons for prioritising

The literature review noted that it was critical to understand the criteria by which the priorities were set, with Nasser, Welch *et al* (2013) stating that this was key to ensuring true and diverse involvement. How this should be done was less clear. In the exercises from Chapter 2 we saw that, for those that did state their criteria, some used preset ones while others had the stakeholders discuss and explore them through the exercise. Viergever *et al* (2010) stated that they should be decided by the participants at the beginning of the exercise and noted that they generally fell into three domains: public health benefit (*should we do it?*), feasibility (*can we do it?*) and cost.

In the Dutch and American exercises discussed in the previous section, the criteria were preset, and were stated to be:

- relevance (severity, prevalence, costs – van Benthem *et al* 2013) / desirability of research (Patel *et al* 2013);

- urgency (van Benthem *et al* 2013) / importance (Patel *et al* 2013);

- impact on the field / society (both);

- and researchability / feasibility (both) / duplication of research (Patel *et al* 2013).

These fit within the domains given by Viergever *et al* (2010), with the first three addressing public health benefit and the last feasibility. However, are these domains enough to ensure true involvement and inclusion of views in a multi-stakeholder exercise?

In this exercise, the criteria for prioritisation were discussed by the stakeholders throughout the exercise. Reviewing Table 26 (Chapter 5, page 160) many of these criteria fit within the Viergever et al (2010) domains. For example, burden of disease, large unmet need and the potential for population health benefit all appear there and fit into Viergever' s domain of public health benefit. Similarly, the feasibility domain featured as part of the exercise in the Expert Forum.

This exercise did introduce other criteria that extend beyond the Viergever domains or those used in the Dutch exercise. These fell into two other areas: individual impact and strategic considerations. For the first of these two,

impact on the health and quality of life of individuals, rather than at the population level, was seen as a reason to both suggest and prioritise ideas throughout the exercise. This individual level of impact as a prioritisation criterion was also recognised in some of the other exercises where criteria were inductively developed. This was the case for exercises that only consulted patients (Tong *et al* 2008) as well as those involving multiple stakeholders (Jagsi *et al* 2012 and Chalkidou *et al* 2009).

The other criterion in this exercise was considering strategy in developing a research agenda. This was seen where priorities were chosen as they would result in a balanced research agenda. Such agendas were seen to provide rapid research gains highly likely to result in return on investment, as well as aspirational longer-term objectives. An example of this was in the reasons for choosing the priorities in the Adult Hearing Loss and Tinnitus small discussion group at the Consensus Conference. There the priority "What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?" was specifically chosen because it was aspirational. Some of the participants felt that the gains from this priority were too far off to be considered as part of a research agenda. After discussion the group was prepared to select this as they felt it was balanced by the more "rapid wins" that could be gained from their priority about hearing aid strategies.

This pragmatic approach has a dimension beyond the Viergever domain of feasibility. It was less about whether the research could be performed, and

more around how to justify higher risk investment that had the potential for transformation.

What this demonstrates is that the more formally recognised criteria against which research ideas are judged, such as those used in the Dutch exercise or the Viergever domains, may not fully reflect the reasons that different stakeholders would use to make their decisions. Although pre-setting criteria may be useful to make sure that important considerations are not overlooked, they may also exclude stakeholders who may have different values. Thus, allowing the stakeholders to discuss and determine their own reasons for prioritising ideas was a strength of this exercise as it avoided this subtle measure of exclusion.

6.3.4 Methodological contributions: using framework analysis to promote inclusion

As was acknowledged in Chapters 1 and 2, research priority setting has been an evolving field. In the additional three years of the updated literature search when this exercise took place, there were seven new overview or methodology papers published in this field, as well as 103 new research priority setting exercises that were in scope of the original search. The large number of the latter would appear to be due to the increase in patient organisations and professional societies conducting James Lind Alliance Priority Setting partnerships to determine research priorities. Yet despite the growing popularity of this methodology, both its originators and users have recognised that it has limitations. Specifically it was felt that the methods used

did not optimise inclusion, especially for marginalised groups (Pollock et al 2014), and that the original meanings of the responses were lost or changed by the need to reformat and reword them into research (PICO format) questions (Uhm et al 2015).

These concerns around inclusion and capturing the meaning or the voice behind the ideas continued in the more recent literature. Fleurance et al (2013) noted the challenges faced in setting research priorities due to the lack of a common language between patients and researchers. Pratt et al (2016) stated that unless there was deep inclusion of stakeholders, rather than simply broad inclusion, there was a risk that research priority setting could amplify inequality. They outlined that the process itself would entrench prejudices by appearing to be inclusive while actually allowing more traditionally respected voices to dominate.

The exercise in this thesis considered these concerns around inclusion. In order to address them, consideration was given to how to design methodology and methods to incorporate all the stakeholder voices. The decision made here was to use qualitative methodology and framework analysis. In this exercise a framework was inductively developed based on the views of the stakeholders gathered through the survey and focus groups, using framework analysis. From the initial literature search this method of analysis had not been used previously for a priority setting exercise, though some of the previous exercises in other fields had used thematic analysis to analyse and

present their results. In the repeated search after the completion of the exercise, two papers discussed the use of frameworks.

One of these papers (Lawlor *et al* 2014) used an analytical framework based on the care pathway to frame research priorities. This was a deductive approach based in Decision Theory, rather than framework analysis using the Ritchie and Spencer (2003) model. The other paper (O'Brien *et al* 2014) used content analytical technique to inductively develop research ideas, though the framework that they used was a pre-existing one that was accepted in their field of research. Neither of these were in ENT, Hearing and Balance care, and neither developed a framework from the data gathered in the exercise.

The approach used for this thesis was flexible and allowed for the organisation of large amounts of information and iterative development between team members, while still maintaining links to the data. It was chosen as it could be used for the inductive development of concepts from stakeholder responses. However, it must be asked if a framework approach was the right way to assess the data?

Through the literature there was tension between remaining pure with regard to the methodological approach and being pragmatic with regard to what was trying to be accomplished with the output of the exercise. Abma (2005) warned against the reductionist trap of turning "experiential knowledge into abstract research themes" and thus losing rich data that could inform future research questions. This was in juxtaposition to the view of Viergever *et al*

(2010) which recognised that themes may be more accessible to those looking to implement the priorities. This could be resolved by trying to capture both, as was done in this exercise, through developing a framework populated with more specific ideas and details. However, even in this approach there were difficulties in determining how granular the research ideas in the framework should be.

In developing the framework in this exercise there was concern about how recognisable this would be to all the stakeholders, and this was reflected in its final form. The development of topics, as well as themes, to create the two-dimensional framework was controversial in the Academic team as there were concerns about their biomedical-centric focus. There was considerable discussion as to whether the topics were as clearly inductively derived from the data as the themes had been, or if this was what the clinical academics on the Academic team expected to see (Chapter 4, page 118). The topics were recognised as useful in that they provided familiarity with the stakeholders who would potentially implement them.

It could be questioned whether this truly reflected stakeholder voices, where different stakeholder categories would not necessarily recognise the biomedical aspect of the framework. This was not highlighted as an issue in the Consensus Conference, although participants were not asked directly about the form of the framework and discussion with them focused on the priorities.

This could be considered a pragmatic though flawed approach in keeping with Viergever *et al's* (2010) suggestion. Thus, what was developed in this exercise was a new structure for UK ENT, Hearing and Balance care developed from stakeholder views that was still recognisable to those that allocate funding.

This thesis is not proposing a new methodology for priority setting. However, it has added to the literature by using a qualitative approach with a variety of methods and framework analysis for the first time. This allowed the incorporation of the views of a range of stakeholders and developed a shared understanding of priorities. By acknowledging the trade-offs between a purely academic approach and the pragmatism needed to develop priorities that would be adopted it has demonstrated the tensions inherent in such a political exercise. As such it provides a case study and outlines these considerations clearly for those designing such studies in the future.

Having considered the contributions of this thesis, its impact on UK research activity is discussed next.

6.4 Impact of this thesis: UK research activity post GENERATE

In the first chapter of this thesis, the research activity in UK ENT, Hearing and Balance care was considered. The aim of this thesis was to develop the research priorities, but the wider aim of the GENERATE project was to

stimulate research and funding in the field. In this section, research activity and funding, particularly that related to the priorities will be considered.

The GENERATE research agenda, based on the priorities developed in this study, was completed in December 2015. The research activity following the launch of the agenda was assessed through examining the UK NIHR Clinical Trials Gateway (https://bepartofresearch.nihr.ac.uk/), and through proactive and reactive discussions with members of the UK ENT, Hearing and Balance care research community. This was not intended to be a formal, exhaustive search, but rather an overview of post-GENERATE activity.

In terms of overall research activity, The NIHR Clinical Research Network (CRN) supported 71 ear nose and throat studies, 25 of which were new studies and recruited 3,798 patients to studies in 2018/19 (NIHR 2019).

Two of the areas that have received the most significant investment were the Adult Hearing Loss and Tinnitus topic, with three of the four priorities being addressed, and Children and Young People. In 2016, the NIHR announced funding for three Biomedical Research Centres (BRCs) for hearing research. This dedicated £15 million to be spent over five years on hearing research for adults and children (Department of Health and Social Care 2016).

Subsequently, these BRCs focused on themes aligned with what had been prioritised. For example, the Manchester BRC started developing a set of core outcomes to measure effectiveness of paediatric auditory devices and

treatments (NIHR Manchester BRC 2017). This related to the priority: *What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?*

Another example was the University College London Hospital (UCLH) BRC which developed a study to design a platform for integrated decision support and holistic management for people who use hearing aids (NIHR UCLH BRC 2019). This addressed: *Which hearing aid strategies work best for adults with hearing loss?* This BRC also supported a trial of a novel drug aimed at restoring hearing in adults with sensorineural hearing loss, which aligns with *What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?*

The Inflammation, Infection and Allergy topic also received funding and initiated research across three of the four priority areas. A national research collaboration across three centres in England received £3.2 million in the form of a Programme Grant. This was to determine current pathways of care for rhinosinusitis, assess the effectiveness of treatment in this area, and then develop new pathways of care (MACRO Programme 2020). This related to the priority: *What are the most clinical and cost-effective treatments for chronic rhinosinusitis?*

Two further priorities were covered in this topic. *Define the different variations of disease that make up rhinosinusitis ("endotypes" and "phenotypes") and develop ways of classifying them based on signs, symptoms and biochemical markers* became the subject of a PhD study (NHS Health Research Authority 2016). *What outcome measures and instruments should we use in primary and secondary care for chronic ENT infections and inflammatory conditions?* informed the development of the DECODE project: DEveloping a Core Outcome set and Diagnostic criteria for acute otitis Externa (Smith 2019).

In addition to increased overall activity, and significant funding for priorities identified in this study, the formation of research collaborations and networks have been noted. In Chapter 1, page 28, Witsell *et al* (2013) noted that the formation of such networks was important to further ENT, Hearing and Balance care research. One of these collaborations, for the rhinosinusitis Programme Grant, has already been mentioned. This combined academic centres, as well as researchers from different backgrounds, such as Health Informatics, Health Economics and Qualitative Researchers, with clinical academics in ENT surgery.

Another national research collaborative was formed following the end of the GENERATE project. This was called INTEGRATE and was a trainee-led research network. Formed in 2016, it started delivering national research projects, including the DECODE project outlined above. It also aimed to provide ENT trainees with research skills to foster their ability to lead their own projects regionally and locally (Smith 2019).

In Chapter 4, page 114, it was noted that during the development of the framework another theme, 'Innovations in Research', had been discussed. This was felt to contain strategies for realising the priorities, rather than research ideas. Developing capacity, establishing collaborations, setting up data-based platforms, and increasing the quality and value of research performed were seen as key strategies. In assessing the research funding and activity after this study, it can be seen that, alongside a number of the priorities being addressed, these strategies have also been realised.

It would be overstated to claim that this study was the cause behind all of this. Nevertheless, all the activities described above had investigators or supervisors who had participated in the exercise or had contacted the author of this thesis to discuss the findings. In particular the INTEGRATE research network was developed following the launch of the research agenda based on this thesis, and its name was chosen to mirror GENERATE. One of the founders of INTEGRATE had been a member of the GENERATE Steering Committee. Considering this, in many ways this study could be considered a description of the zeitgeist at the time immediately preceding the increased activity. Even if this study's role has been only to provide clarity in that description, this would still be a considerable impact on understanding how research in ENT, Hearing and Balance care developed. However, given the wide engagement of the research community in this exercise and specifically those involved in the projects outlined this RPSE has been influential in shaping the research activity, rather than simply describing it.

6.5 Future research, policy and practical considerations

This study of UK ENT, Hearing and Balance care research priorities has highlighted several areas for further research, as well as policy and practical considerations for the specialties.

One of the Viergever et al (2010) criteria for good practice was to evaluate the priorities after they had been determined. This was seen as having three components: reviewing the process, periodically reviewing the priorities and conducting an impact analysis. In the more recent literature, Bryant et al (2014) noted the lack of assessment by exercises on policy, practice or improvement on outcomes. They did note that what had been assessed was participant satisfaction with the process and their perception of the discussions in the exercise. They noted that all of the latter showed positive results.

This lack of impact analysis may be due to the different approaches taken to priority setting and the reasons behind these different approaches. If the sole purpose of the exercise was to increase funding for specific projects, then it may be that assessing how many of the priorities were funded for research after a set period of time is a sufficient metric. There are potential confounders to this approach, such as other drivers for the funding, which would need consideration. If, however the part of the purpose of the exercise is to promote

the culture of research in the specialty or field, this would be harder to assess. Research into outcome evaluation in general and for different approaches would be valuable to the field of research priority setting.

In this thesis, the process that was developed has been reviewed as per the Viergever *et al* (2010) criteria. Moving forward, redetermining the priorities and more formally assessing the impact of the exercise based on the different considerations above would be informative and an area of potential future study.

Another area that would benefit from ongoing research is understanding the criteria that different stakeholders use to determine priorities. As was seen in the literature review in Chapter 2, of the 113 exercises identified only 28 reported the criteria used to determine the priorities. Of these, 20 used pre-set criteria. A study of inductively derived criteria from different stakeholder categories would be valuable. It would inform funding bodies and the designers of exercises with pre-set criteria of considerations that may not have been used traditionally in allocating research funding.

The more recent literature has shown some work that has been done in this area, with a study comparing the preferences of researchers and laypeople regarding the outcomes of biomedical research (Miller et al 2013). However, even this study used pre-set criteria rather than exploring different perspectives as to what constituted value in research. If inclusion is seen as important in setting priorities, then re-examining what is traditionally seen as

valuable in research is important. Otherwise, despite including "non-traditional" stakeholders, there is a risk that their views will be marginalised.

This thesis has also highlighted policy and practical considerations to be considered when developing and implementing a research agenda based on the priorities identified. In Chapter 4, pages 114 – 115, the ideas that stakeholders proposed to accomplish this were outlined. As had been described above, some of these suggestions have been realised.

There is still more that can be done. The policy levers to be considered for implementing these changes lie with the professional bodies developing research and the educational bodies developing clinicians and researchers, including lay researchers. By placing emphasis on the value rather than the volume of research being done, and by improving training in research methodology and methods, better quality research done by people with a range of backgrounds is possible, Furthermore, incentivising and supporting junior clinicians and patients to undertake research on a national level beyond that provided by INTEGRATE will promote improved research practices going forward. Practically this would mean engaging with employers and trainers to ensure that consistent professional time could be dedicated to research, without clinical interruptions.

Creating more platforms would also require dedicated resource and would be improved by developing them collaboratively with a range of stakeholders so that they were fit for purpose. Specifically, involving patients in designing data

collection platforms so that they capture outcomes that are meaningful to them through modalities that are practical for them would be important.

This discussion has also considered the impact on research activity. In Chapter 1, it was stated that the wider GENERATE project was been designed to address both the general concerns over developing a high value research agenda and the specific concerns about the state of evidence for commissioning ENT, Hearing and Balance care. The output of that project was seeking to influence allocation both of research resources and, ultimately, healthcare resources through the eventual evidence derived from the priorities.

Clearly there are many drivers behind cutting funding to interventions and services, and increasing the evidence base demonstrating benefit of these in ENT, Hearing and Balance care will not guarantee desired levels of funding. In moving forward, the stakeholders will need to combine efforts to negotiate and design services based on the evidence generated.

6.6 Study design and the quality parameters: strengths and limitations

The strengths of this study have been shown through this thesis and the previous sections in this Chapter. These have been considered through the lens of the three quality parameters highlighted in the literature review, as well as through considering how the design fulfilled Viergever *et al's* (2010) checklist. These will now be discussed further, along with the limitations.

Transparency: From the literature, transparency was seen as crucial to research priority setting exercises. This has been interpreted as transparent reporting of the exercise (Viergever *et al* 2010; Montorzi *et al* 2013; Whitlock *et al* 2010). Although this exercise was designed so that there was transparency throughout, what was not apparent in this thesis was the transparency with which this reporting was disseminated back to stakeholders. As outlined in Chapter 1, this thesis was part of a broader initiative to promote engagement in research in ENT, Hearing and Balance care. The objectives of this thesis were to design a research priority setting exercise and determine the research priorities according to the stakeholders, therefore the full dissemination activities have not been captured here, though a copy of the final report may be found in Appendix 14, page 361.

Inclusion: In this thesis inclusion was considered from two perspectives:

- spread of respondents and participants through the different stages of the process (*did they have the opportunity to participate?*);
- how the methodology and methods used both promoted inclusion of different voices and prevented subtle means of exclusion (*did they have the opportunity to be heard?*).

Stakeholder spread: This exercise was designed to be a multi-stakeholder exercise. At each step of the process efforts were made to ensure that a range (rather than simply a large number) of stakeholders was consulted. However, there was potential underrepresentation of some categories. In the

survey there was poor response from the Industry category and concern that 'unorganised' patients (as per Corner *et al* 2007), carers, and healthcare professionals beyond ENT surgeons had not been fully heard. To rectify this there was targeted outreach to these stakeholders in the focus groups. With regard to Industry, participants from the pharmaceutical and medical device industries were present at a separate group, which meant that their views were incorporated. A limitation here was that there were further industries, such as private providers of healthcare, that could have participated.

There were focus groups of non-ENT healthcare professionals who dealt with relevant conditions in different settings. The limitation here was that a full range of mixed healthcare professionals was not recruited. For example, no Speech and Language therapists were present at Focus Group 1, which weakened the claim to truly present ideas from a mix of professionals. However, it is worth noting that there had been a broad spread of clinician respondents to the survey even if numbers were lower, therefore this limitation was accepted.

The range of stakeholders involved in the Expert Forum was necessarily limited by the role of the group. Nevertheless, within these confines a range of expert stakeholders across different therapy areas and disciplines was recruited. This provided a range of perspectives and expertise beyond the scope previously described in ENT, Hearing and Balance exercises.

The Consensus Conference had participants from all stakeholder categories, with the exception of Industry. This was at the suggestion of the Steering Committee and the Expert Forum, in order that the final priorities chosen were not seen to be influenced by specific companies or interests. This could be seen to be limitation in an exercise designed to include all stakeholders. However, this moved participation in UK ENT, Hearing and Balance exercises from previous targeted exclusion to targeted inclusion of Industry voices. This was a fine balance between fulfilling the research question and being pragmatic about the adoptability of the final priorities by the stakeholders.

Inclusion through methodology and methods: The main concern raised in the literature was that patients and carers were at risk of poor inclusion especially from those considered 'unorganised.' Targeted strategies to promote inclusion were a strength of this exercise. One of the particular strengths was the use of framework analysis to promote full inclusion of stakeholders, which has been noted as a methodological contribution of this thesis. Why this was a strength will now be considered further.

When developing the research ideas to populate the framework it was challenging to combine concepts from multiple stakeholder responses while keeping enough detail so that research projects could be designed from the results. This was specifically commented on in the Expert Forum (Chapter 5, page 189).

As an added challenge, the more granular the output, the more likely it was to have devolved from the detail provided by the views of fewer stakeholders. This was seen where some of the stakeholders had detailed technical knowledge that could be easily translated into traditional biomedical research questions. This ran the risk of giving more weight to responses from these stakeholders.

Placing such responses into the framework alongside more experiential ones from other stakeholders helped alleviate this. It put the more traditionally recognisable ideas into the context developed from the different stakeholders and helped with combining related ideas from different sources. This emphasised the strength in using this approach to analysis. It provided a framework for shared understanding amongst stakeholders, which was advocated in the literature (Abma 2005).

Further strengths of this exercises were the checks used to determine if there actually had been inclusiveness. In order to check that this approach had resulted in ideas being generated from a range of respondents, the priorities were mapped back to the stakeholders who had contributed to the idea. Both the list of ideas that were collated at the end of the Expert Forum and the final priorities were also mapped across the Framework. This was to see if they were spread across the themes. As the ideas had been discussed in groups that correlated to the topics clearly there would be a spread of priorities across them. However, the themes had been developed from the detailed analysis of the data and not influenced by the pragmatic considerations that

resulted in the development of the topics. This was a further check for bias from the Expert Forum and Consensus Conference.

Looking at the spread of ideas through the framework acted as a proxy measure of exclusion of stakeholder voices. The rationale for this was that the framework had been rigorously developed from the whole of the data; if themes were neglected it would indicate that ideas weighty enough in the data to form these themes had been excluded by the narrower group of stakeholders prioritising them. Thus, these two approaches to mapping were complimentary: mapping priorities back to stakeholders assessed if all stakeholders had had input into the priorities. Mapping back across the framework ensured that key ideas that mattered to different groups of stakeholders were not excluded.

As can be seen from Table 24 (Chapter 5, page 150), the priorities were developed from ideas from all stakeholder categories. Each idea derived from more than one category, though not every stakeholder category contributed to each idea. All of the chosen priorities had their origins in data from patients and healthcare professionals, and each had more than one type of healthcare professional respondent contributing to the idea. This meant that these were the categories that had the greatest contribution to the final priorities. This is unsurprising due to the fact that the greatest number of survey responses came from these groups, therefore their contribution was more likely due to this than any particular prioritisation bias.

Reviewing the mapping of the priorities across the themes, it can be seen that they appear to have clustered in the "Clinical and Cost-effectiveness" theme, with just over a half (13 of the final 21 priorities) originating from there. From Table 23 (Chapter 5, page 138) and we can see that just over a third of the research ideas presented to the Consensus Conference were from this theme (36 out of 98 ideas). This can be compared to the number of ideas from "Understanding health, illness and disease" that were prioritised. Only two ideas from this theme were prioritised, although 24 out of 98 ideas presented to the Consensus Conference originated from there. The spread of the final priorities across the rest of the themes is similar to that of the research ideas presented to the Conference.

Although this gave rise to concern that there was bias towards this particular theme in the Consensus Conference it is unsurprising on reflection. The exercise as a whole produced a larger proportion of ideas under "Clinical and Cost-effectiveness" than any of the other themes. The original scope of the exercise was to consider ideas in clinical and health services research, and the question asked in the survey was designed around this. Thus, it encouraged these responses, resulting in a weighting toward this theme, which was a potential weakness of the survey design. The greater anomaly was the identification of the "Understanding health, illness and disease" theme and the relatively large proportion of questions there, when the survey did not ask or provide prompts for ideas relating to this. The fact that this was an identified theme despite the potential skew of the survey demonstrates that the stakeholder views and voices were not unduly limited by this weakness.

One area of potential exclusion of stakeholder views and voices was seen in the theme of "Creating effective communication." There were no ideas that appeared in this theme in the final priorities. However, as was seen in Chapter 5, page 193, the discussions around the final choices were more nuanced than this would indicate, and ideas from this theme were seriously considered. Interestingly, the academic analysis would have placed the "Balance Problem" priority concerning self-management in this theme. This would have meant that all the themes were covered in the final priorities. However, as was shown in the analysis of that group's discussion, the participants placed it under the "Promoting Knowledge and Learning" theme. The tension between keeping these two themes separate or combining them was present throughout the development of the Framework (see Chapter 4, pages 110 - 111). Therefore, the exclusion may reflect more on the final decision to separate the themes in the framework than on exclusion of ideas in the prioritisation process.

There were also limitations in inclusion in this study. Although all the planned focus groups occurred, they were of variable size. Two of the groups were outside the aimed size of six to ten members. Due to resource constraints and the difficulty in recruiting participants of a similar calibre a second time, these limitations had to be accepted, especially as they provided ideas from stakeholder categories underrepresented in the survey.

In the Expert Forum it was felt that the power imbalance between academic experts and 'unorganised' patients would be too great to bridge, and that for there to be meaningful deliberation there needed to be deliberate recruitment of patients experienced in research (as per Campbell 2010 and Corner et al 2007). Thus, the patient that was recruited had significant university PPI and hospital board experience. This was felt to be a strength rather than a limitation at this stage of the exercise. However, the aim was to recruit two expert patients, in keeping with best practice (INVOLVE 2012). Unfortunately, one of these could not attend, which was a limitation in this step.

From this discussion it can be seen that although there were limitations to the outputs and inclusion in some of the steps of the exercise, on balance they would appear to be based on the views and voices of a range of stakeholders across the categories, without obvious exclusion.

Credibility: In Chapter 2 credibility was seen to relate to a robust process with triangulation of methods, the checking of results with stakeholders and a clear audit trail from the data to the priorities. All of this considers whether the outputs are reflective of the stakeholder views and resonate with them (*were the stakeholders heard and do they recognise that they were heard?*).

In this exercise there was triangulation of methods in different stages of the process, including using multiple researchers to analyse the data. One potential limitation here was that all the analysts were researchers. This had

the advantage that they were experienced in analysing data but having a lay or patient analyst may have enhanced credibility even further.

There were several points during the different stages of the process when results were checked with stakeholders, most notably with members of the Steering Committee, Expert Forum and Consensus Conference. In some cases this resulted in altering the data analysis. For example, in Table 26 Chapter 5, page 160, the priority "How can patients be empowered to self-manage their balance problems and how does this impact on their outcomes?" was refined from other ideas and placed in a different theme in the framework. Participants in that group felt that this reflected stakeholder, particularly patient, ideas more accurately. In this instance it was viewed as acceptable as it enhanced the credibility of the final priorities by checking that they resonated and provided a balance to the lack of a patient researcher in the Academic Team.

The good practice checklist: The Viergever *et al* (2010) checklist was used during the design of this study to try to ensure good practice. It fulfilled most of the criteria outlined, though it does also highlight some potential limitations.

One of these was in considering what information to gather to inform the exercise. Viergever *et al* (2010) stated that this was about knowing where the gaps in current knowledge are and suggested that literature reviews may be needed. This approach was not taken in this exercise; rather an Expert Forum was used to discuss whether the ideas gathered from stakeholders had been

or were being sufficiently addressed. The reason for the decision was that even if research existed in the area under consideration, it may not be considered adequate and may not affect practice. One of the points of discussion in the Steering Committee around this area was that even when the academic community may have felt that research had addressed a specific question, there still seemed to be doubt in that area (often from clinicians) and the question arose: how can you ever tell if the research has been enough?

This is a question that has been seen repeatedly in ENT, Hearing and Balance care. As stated in Chapter 1 (page 18), research considering the use of ventilation tubes or the efficacy of tonsillectomy has been considered to be extensive, yet even so the use of the evidence generated has been inconsistent. Therefore, the more "traditional" methods of assessing evidence, such as literature reviews, was reconsidered. The use of clinical experts, with roles in management and local policy, alongside patients and traditional academics, was felt to give an insight into clinical and resource allocation needs from the evidence. This was felt to be a strength of the exercise. It was recognised that there was a potential concern that using experts introduced a degree of subjectivity and personal bias; this was raised by the experts in the forum themselves. One of the requests from the participants was that the results should be traced back to the original data to make sure that this could be audited; this was done as has been outlined in Chapters 4 and 5.

Another potential limitation from the checklist was in regard for plans for implementation of the results. In considering this, it helps to understand what Viergever *et al* (2010) meant by such plans. Here they recognised that priorities are often not set by those who must implement them. To counteract this, they suggested that careful stakeholder mapping is done prior to starting the exercise, and that policymakers and funders are specifically considered as stakeholders. This was clearly done in the present exercise. Another recommendation was the classification of priorities into themes to make them more relatable to those implementing or funding them, which has been done in this case.

6.7 Critical reflections

The discussion above has focused on the impact of this study on the wider body of knowledge, both for priority setting and with regard to the specific priorities for ENT, Hearing and Balance care. The framework, ideas and priorities developed in this exercise are not technically perfect. Flaws and limitations have been discussed above. Pragmatic choices were made to develop the framework where others may have chosen methodological purity. The priorities in this study themselves vary between broader and more defined ideas and could be seen therefore as inconsistent. However, when viewed alongside other priorities found in the literature these may be seen not as fatal flaws, but rather the consequences of the choice to involve stakeholders with different views and expertise. Seen through this lens that inconsistency is actually a strength as it indicates that the research-

experienced stakeholders with greater technical knowledge have not dominated the other voices.

Ultimately, it is the stakeholders themselves who could best determine if they had felt included and if their views and voices had been heard. This was not a question that was directly asked of stakeholders, but participants in the Consensus Conference did comment about whether they felt that they were able to participate, that their perspectives had been included and whether they had had an impact.

Not everyone present felt that the framework fully incorporated their priorities. This was seen in the Children and Young People's small discussion group. From their perspective, having children and young people as a separate topic was disadvantageous and marginalising, putting them "at the bottom of the list" and narrowing their priority options compared to adults, who could choose priorities from across the range of topics (Chapter 5, page 146). However, by exploring this in discussion and being able to explain this to the larger group, they were able to ensure that some of the adult priorities could include the younger population. This meant that they were heard and were able to directly impact on the final priorities.

Other responses were very positive about their ability to engage in the exercise. Participants in the Inflammation, Infection and Allergy group noted the harmony in the group and felt that the different stakeholders were "in step," with one participant explaining that in their previous experience of

ranking ideas that different stakeholders started out seeing things differently, which they hadn't experienced in this exercise (Chapter 5, page 140). This indicates that they felt that there was a shared understanding between stakeholders by the final step of the process.

Participants were also clear in all the groups that there had been considerable debate and discussion. It can be seen in the final priorities that various stakeholders had effectively swayed opinion to their viewpoint in the groups, but this was not obviously favouring one particular stakeholder category. In the Head, Neck and Airway group patient participants strongly advocated for "What is the optimal management for adults with voice problems?" and convinced more reluctant clinicians to prioritise this. This demonstrates both directly through comments by participants and by reviewing the group discussions, that different stakeholders felt engaged and that they had the ability to be heard in the determination of the priorities.

Thus, although the analysis was not seen as perfect by all the stakeholders, all the groups reported that they had had active discussions and the facilitators ensured that everyone was enabled to participate. In the political environment of priority setting, perfection for all parties is unrealistic; and as Campbell (2010) stated, the opportunity for deliberation with the ability to express dissent and have it acknowledged is, possibly, more important than the final output.

Considering the demonstrated quality of the process, with transparency, inclusion and credibility, as well as the recognised ability of participants to deliberate, the outputs, though imperfect, may be said to be the research priorities that reflect the views and voices of the stakeholders in this exercise.

Figure 23 Critical Reflections: The voice of the researcher

In order that the critical reflections are complete, a final stakeholder voice needs to be captured: my voice as researcher. Of all the quiet voices in this study, I recognise that mine is potentially the most powerful, the loudest. I have shaped the research, guided its direction and played a central role in interpreting all the other voices. When I started writing this thesis, I wrote in the first person. But I found my voice was too loud throughout and interrupted the clarity of the work. Here I will step out of the formal third voice adopted in the thesis and provide critical reflections on my own role in this research and the impact I have had throughout.

At the time that I undertook this research I was a practicing ENT registrar nearing the end of my surgical training. Just prior to starting this study, I had completed a fellowship in healthcare management and clinical leadership, where I had been involved in developing consensus-based commissioning pathways in direct response to the proposed service restrictions suggested by the "Croydon list" of low-priority treatments. I also became involved in developing a NICE-approved consensus process for developing commissioning guidance, which was adopted by the Federation of Surgical Specialty Associations. This background in working with consensus-based approaches and my work in service commissioning led to my interest in this research and had an impact on its development.

Following my fellowship, I was aware of the need to develop the evidence base underlying services. After discussion with one of the professors who formed part of my supervisory team, I proposed developing an RPSE to the ENT, Hearing and Balance professional bodies. I secured their agreement and raised partial funding for the study through them. The consensus-based approach agreed by the Steering Committee, and the need for multi-stakeholder involvement, was influenced by my background. I presented them with the different potential approaches to this work but, based on my experience, advocated strongly for the ones that were ultimately agreed.

I developed all the data collection and prioritisation instruments and facilitated the focus groups at the data gathering stage. As Researcher 1, I immersed myself fully in the data to lead on their interpretation. I was concerned that my clinical background and fellowship experiences would bias my analysis in favour of the stakeholders with whom I had a shared background. To mitigate this, I recruited an Academic Team that had varied expertise. They balanced different types of clinical experience with non-clinical academics from different disciplines. Some of the more heated academic discussions were around moving away from a biomedical model in the Framework. I found this very difficult in my analysis, though the more I developed as a researcher through this process, the more I was able to challenge my innate assumptions about how research "should" be presented and was able to reflect this in my interpretations.

In order to further mitigate my own potential bias influencing the outputs, I did not facilitate any of the groups in the prioritisation stages of the study, instead acting and taking notes as an observer. This gave me a further opportunity to observe how different stakeholders responded to the analysis presented to them, without taking part in the discussion myself. This provided useful reflection to shape further analysis through these stages, and to challenge my previous choices of concepts and words, both personally and in the Academic Team meetings.

While writing this thesis I have had a chance to reflect further on my role as both a junior surgeon leading in a profession-wide exercise, and as a junior researcher taking a lead role in an experienced Academic Team. Having my own area of expertise through my fellowship experience was important in my ability to express my ideas confidently. As I grew in research experience, I recognise that I relied less on the perspective and standing of my past. I was able to listen more clearly both to the perspectives of the wider team and to the voices of the stakeholders involved. I was struck by the trust that was placed in me by all the stakeholders and communities involved despite my lack of seniority. I also recognise that the fact that I was junior was an advantage in that I was perceived to have fewer entrenched interests than someone established in their career. This makes the acknowledgment of my own background and role in this study of added importance, so that it is clear that I did bring my own assumptions and experiences to this endeavour.

6.8 Conclusion

Research priority setting has become an increasingly recognised way of mobilising research activity by engaging stakeholders to come to a consensus about shared priorities. This thesis has presented the first UK research priority setting exercise for ENT, Hearing and Balance care based on the views and voices of stakeholders, as well as contributing to the growing body of evidence on the process of setting priorities. GENERATE, the exercise that is the study of this thesis, has both been a part of and contributed to the resurgence of clinical research in ENT, Hearing and Balance care. Whether it will result in the ongoing design and funding of patient-centric services remains to be seen, and will rely on the next generation of researchers to continue the momentum and maintain the networks it has inspired.

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Appendices

Appendix 1: Search strategy

1 exp Biomedical Research/ (182917)
2 (health$ adj3 research$).mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (57163)
3 (clinical$ adj3 research$).mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (69193)
4 1 or 2 or 3 (256596)
5 exp Health Priorities/ (8826)
6 priorit$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (66928)
7 prioritise*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (1298)
8 prioritize*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (7151)
9 prioritisation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword
heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (657)
10 prioritization.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword
heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (3334)
11 prioritization.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword
heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (3334)
12 agenda$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (12395)
13 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 (77556)
14 4 and 13 (8262)
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word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (1620969)
16 consensus.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (112726)

17 method$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (4101823)
18 approach$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (1048134)
19 design$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (1341258)
20 15 or 16 or 17 or 18 or 19 (6687770)
21 14 and 20 (5341)
22 (priorit$ adj2 setting).mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (2125)
23 (agenda$ adj2 setting).mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (338)
24 exercise$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (250131)
25 22 or 23 (2459)
26 24 and 25 (145)
27 "priority setting exercise$".mp. [mp=title, abstract, original title, name of substance word, subject heading
word, keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (53)
28 4 and 26 (46)
29 (research adj3 prioritisation$).mp. [mp=title, abstract, original title, name of substance word, subject heading
word, keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (30)
30 (research adj3 prioritization$).mp. [mp=title, abstract, original title, name of substance word, subject heading
word, keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (104)
31 (research adj3 agenda$).mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (3957)
32 (research adj3 priorit$).mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (4900)
33 29 or 30 or 31 or 32 (8458)
34 20 and 33 (5085)

35 ("research agenda$" adj3 priorit$).mp. [mp=title, abstract, original title, name of substance word, subject
heading word, keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique
identifier] (86)

36 29 or 30 or 32 or 35 (4909)

37 20 and 36 (2989)

38 limit 37 to (english language and yr="1990 -Current") (2723)

39 health$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (2400005)

40 healthcare$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword
heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (104989)

41 clinical$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (3027879)

42 biomedical.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading
word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (106270)

43 39 or 40 or 41 or 42 (5015958)

44 38 and 43 (2100)

45 "health service$".mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword
heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (329839)

46 "healthcare service$".mp. [mp=title, abstract, original title, name of substance word, subject heading word,
keyword heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier] (3477)

47 40 or 41 or 42 or 45 or 46 (3453345)

48 38 and 47 (1413)

Appendix 2: Scoping Review Update

<u>Aim and Background</u>:

The aim of this further literature review was to update the scoping review from Chapter 2. The review in that chapter was used to develop the methodology for the research priority setting exercise presented in this thesis. As the exercise was conceived and developed between 2012 and 2013, with it running until December 2015, it was recognised that it was likely that the literature had developed during this period. To consider the contribution of this thesis to research it was decided that an update was needed to further inform the discussion chapter.

<u>Methods:</u>

The method used for the scoping review in Chapter 2 was repeated, using the search period from 1 April 2013 to 30 April 2016 in the MEDLINE, EMBASE and HMIC databases. This period was selected as it coincided with the time during and immediately after this study was performed and was therefore the relevant timeframe to consider the contribution made by this research. After screening of titles and abstracts, the same inclusion and exclusion criteria were applied to review the full texts. Data extraction was again done using Microsoft Excel, in the same format as the first review.

<u>Findings:</u>

109 papers were included in the updated review (see figure 24 below).

ENT, Hearing and Balance RPSE: Two more recent exercises were found in the updated literature search. Details of these are found in Table 28. One of these was a topic specific UK exercise, using Priority Setting Partnership (PSP) Methodology, and was focused on research priorities for mild to moderate hearing loss in adults (Henshaw et al 2015).

The second was an American exercise to determine the priorities for diagnosis and treatment of obstructive sleep apnoea (Patel et al 2013). Following completion of a previous comparative effectiveness review, a panel representing six stakeholder categories nominated, discussed and prioritised future research needs topics.

Methodology papers: 7 papers were identified. These are summarised in Table 29.

RPSE papers: A further 100 exercises were identified. Of these, 23 used PSP methodology, 20 used the Delphi method and two used the Dialogue Model. A further two used qualitative methodology, two used mixed methodology and one specified a participatory approach. The remaining 50 used their own approach, using similar methods to those observed in Chapter 2.

Discussion:

Please see Chapter 6 for discussion relevant to this study.

Database Search:
(MEDLINE, EMBASE, HMIC)
705

Duplicates
42

662

463 publications excluded on title and abstract

200

101 publications excluded on full text

Selected publications n=99

Other publications identified through references and by discussion with experts (n=11)

109 publications included
102 RPSE (2 ENTHB, 100 other); 7 overviews
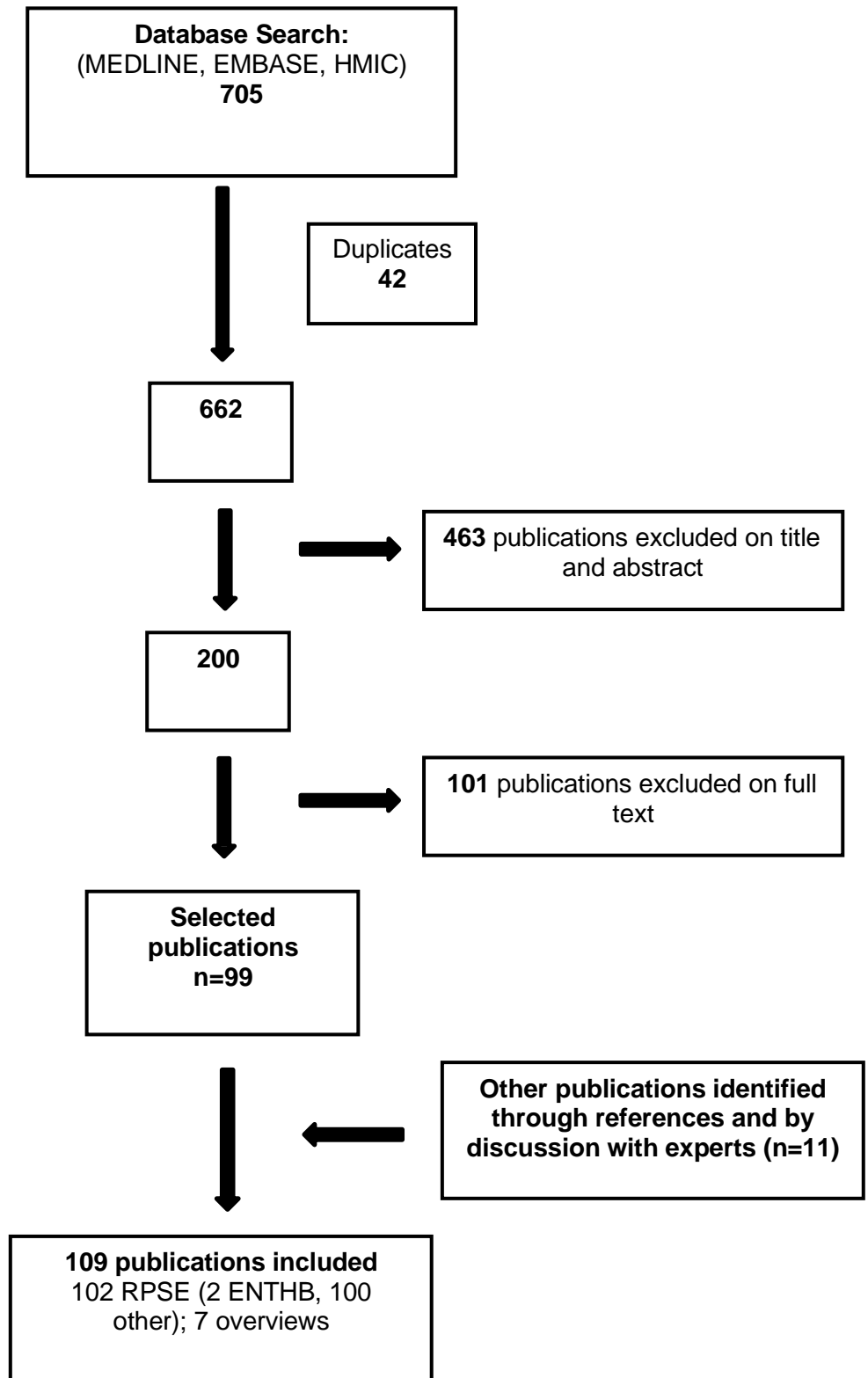
Figure 24 Updated review PRISMA flow diagram

RPSE	Scope	Methodology	Stakeholders	Priorities
Patel, K. *et al* (2013) High priority future research needs for obstructive sleep apnea (OSA) diagnosis and treatment.	USA OSA	Stakeholder panel nominated, discussed and prioritised future research needs based on a completed comparative effectiveness review	Patients and public Providers Health care purchasers Payers Policymakers Principal investigators	Nine highest priorities: Cost-effectiveness of management strategies Defining age- and sex-specific criteria for OSA Evaluating routine preoperative screening for OSA Evaluating involvement of a sleep medicine specialist in diagnosis of OSA Evaluating clinical prediction rules Assessing effect of treating sleep disordered breathing and long-term clinical outcomes Comparing treatments for patients who do not tolerate positive airway pressure Evaluating strategies to improve treatment compliance Evaluating association between OSA severity and long-term clinical outcomes
Henshaw, H. *et al* (2015) Research priorities for mild-to-moderate hearing loss in adults.	UK Mild – moderate hearing loss in adults	James Lind Alliance Priority Setting Methodology	Patients Carers Clinicians	Top ten priorities: What adverse effects are associated with not treating mild to moderate hearing loss in adults? Does the early fitting of hearing aids result in increased patient benefit and/or improved cost-effectiveness of the service? Does the early fitting of hearing aids slow the rate of cognitive decline? What are the reasons for low hearing uptake, use and adherence? Can new technologies replace hearing aids? Can stem cell therapy offer a cure for mild to moderate hearing loss in adults? Does early identification, diagnosis and treatment of mild to moderate hearing loss prevent further deterioration of hearing? Could new developments to digital hearing aids offer improved speech perception in noisy environments? How realistic are hearing tests for assessing the everyday hearing abilities of adults with mild to moderate hearing loss? Could the use of real world sounds to help program hearing aids in clinic (rather than tones or bleeps) improve hearing aid effectiveness?

Table 28 RPSEs for ENT, Hearing and Balance care from updated review

Authors and Paper	Objective	Method	Key knowledge summary	Indicators of quality
Bryant et al (2014)	Develop and examine "a methodology for health research agenda setting processes grounded in the notion of participation as dialogue."	Review and discussion of case studies	The Dialogue Model: six-phase approach grounded in participatory and interactive approaches. Emphasises mutual learning by means of ongoing dialogue and the development of shared action agendas supported by the different stakeholders.	Credibility: participants should recognise the results; enhanced through member check and triangulation. Fairness: enable relevant stakeholders to participate in the process; visibly include their voice.
Chalmers et al (2014)	To develop recommendation for how to reduce waste when research priorities are set.	Expert report with analysis of clinical study reports	More research on research is needed for successful replication. Research funders should be transparent in how and why they fund research Funding proposals should be justified by systematic reviews. Make information about research in progress available and encourage collaboration.	Did not outline beyond knowledge summary.
Fleurance et al (2013)	To describe PCORI's approach to meaningfully integrate the patient voice into research process.	Expert report	Eliciting research questions directly from patients, their caregivers, and their clinicians and asking them what matters to them in their daily lives have the potential to focus clinical research activity on more practical, patient-centered questions. Lack of common language is a challenge.	Deliberative processes which encourage participation and help overcome stakeholder wariness.
Guise *et al* (2013)	To review the current practices for engaging stakeholders in prioritising research.	Literature review and semi-structured interviews	Explicit and consistent use of terminology about stakeholders was absent. Research organisations had difficulties engaging the public and policy makers. Engagement checklist developed.	Use of in-person techniques to generate and clarify ideas. Early identification and engagement of stakeholders in the process. Credibility. Use of multiple methods.
Miller *et al* (2013)	To assess if researchers and the public assess research outcomes equally.	National survey and discrete choice experiment	Citizens and researchers prioritise the same outcomes for basic biomedical research: traditional scientific outcomes rather than economic returns. Has implications for how academic medicine assigns incentives and value to basic health research.	Joint contribution of researcher and the public.

Pollock *et al* (2014)	To develop and use a new model of engagement for patients and clinicians in research priority setting.	Development of engagement model	Reliance on standard surveys may result in poor and unrepresentative involvement of patients, thereby favouring the views of health professionals.	Use of different methods for engagement with marginalized groups; face to face engagement may be preferred.
Pratt *et al* (2016)	To develop a model of deep inclusion.	Development of conceptual model	Justice requires national health research priority-setting to promote health equity. A wide range of stakeholders attains shallow inclusion in research priority-setting. Deep inclusion demands breadth, qualitative equality, and non-elite participation. Achieving all three dimensions is necessary for priority-setting to advance equity.	Equity and deep inclusion.

Table 29 Table of overview and methodology papers for updated review

Appendix 3: Summary table of RPSE publications excluding ENT, Hearing and Balance exercises from initial scoping review

Publication	Topic	Participants	Methodology/Process/Methods (Bold indicates methodology)
Abma *et al* (2005)	Spinal cord injury	Patients, doctors, researchers	**Dialogue Model**
American Society of Nephrology (2005)	Nephrology renal research	Experts, including industry	Expert panel, working groups
Annells *et al* (2005)	District nursing	District nurses	Delphi method
Banfield *et al* (2011)	Mental health	Patients	**Mixed methodology**
Barnato *et al* (2007)	Cancer care (patient decision aids)	Symposium members (not specified)	Expert panel, facilitated discussion and vote.
Bartosh *et al* (2008)	Paediatric solid organ transplantation	Clinicians, researchers, industry, government	Expert working groups
Batchelor *et al* (2013)	Eczema	Patients, carers, health care professionals	**Priority setting partnership**
Bayley *et al* (2004)	Emergency nursing	Nurses	Delphi method
Bedos *et al* (2004)	Dentistry	Clinical dentists	Survey
Blum *et al* (2012)	Developmental-behavioural paediatrics	Paediatricians, psychologists, parents	Delphi method
Boon *et al* (2006)	Complementary and alternative medicine	Researchers, practitioners, policy makers and funders	Modified Delphi process: to identify priority research areas Advisory Board consultation: determine priority topics from Delphi round Prioritisation and strategic planning meetings: identify and prioritise specific research questions and projects within the identified priority research areas
Bousvaros *et al* (2006)	Paediatric inflammatory bowel disease	Multidisciplinary researchers	Focus groups: identified knowledge gaps and recommended research projects. Expert conference: participants considered importance of proposed research items and feasibility in terms of manpower and resources available, then assigned the highest priorities to 5 areas.
Broerse *et al* (2010)	Burn research	Healthcare professionals and patients	**Dialogue Model**
Brown *et al* (2006)	Health and learning disabilities	Clinicians, health service managers, allied health professionals, independent sector, social services, higher education, family carers, service user.	Facilitated focus group design was employed at a conference event

Buckley *et al* (2010)	Urinary incontinence	Clinicians and patient organisations.	**Priority setting partnership**
Burt *et al* (2009)	Colorectal surgery	Colorectal surgeons, academics, basic and clinical scientists	Modified Delphi method
Butler *et al* (2009)	Midwifery	Midwives, consumers of maternity services.	Delphi survey
Byrne *et al* (2008)	Childhood obesity	Research groups	Delphi method
Cairns *et al* (2010)	Emergency trauma care	Basic, translational, and clinical Researchers in acute injury	Expert roundtable
Caron-Flinterman *et al* (2005)	Asthma and COPD	Patients	**Participatory:** focus groups and questionnaire
Carpenter *et al* (2011)	Geriatric Emergency care quality indicators	Physicians, nurses, social workers	Delphi surveys and expert audience consensus at 2 conferences
Chalkidou *et al* (2009)	Coronary artery disease	Representatives from hospitals, payers, product manufacturers, clinicians, researchers, consumers, and government agencies	Workgroup using informal nominal group prioritisation process based on an explicit set of criteria pre-agreed by the group
Chesney *et al* (2006)	Paediatric chronic kidney disease	Experts	Task force with expert panel
Chestnutt *et al* (2000)	Dental public health	Dental public health consultants, non-consultant members of oral health steering group	Delphi method
Cheyne (2013)	Maternity care	Maternity service users	**Participatory:** Modified nominal group technique with rapid literature review
Christensen *et al* (2013)	Mental health	Researchers, Government representatives, mental health consumer groups, carer groups	Online survey
Clavisi *et al* (2013)	Traumatic brain injury	Clinicians, researchers, managers, and patient/carer representatives	**Mixed methodology:** questions generated through scoping meeting, literature search and workshop using nominal group process, plus structured online survey asking for questions structured in the PICO format (generally got idea fragments, so coded and thematically analysed). Prioritization by online survey to rank in 3 domains. Existing research mapped to identify evidence gaps. Priorities for investment set through multistakeholder forum with facilitated group discussions. Smaller targeted workshops then focused on specific issues.
Clinton-McHarg *et al* (2010)	Adolescent and young adult	Patients, carers, multidisciplinary	Value-weighting approach with modified Delphi approach and

		haematological cancer	researchers and clinicians	resource allocation questionnaire
Clohan *et al* (2007)	Post-acute rehabilitation	Government agencies, private insurers, professional organisations, providers of rehabilitation services, patients, health researchers	Symposium with round table discussions and break out sessions
Coleman *et al* (2008)	Metastatic bone cancer	Not specified	Multidisciplinary consensus conference
Corner *et al* (2007)	Cancer	'Unorganised' cancer patients	**Exploratory, qualitative approach**
Corona *et al* (2013)	Scoliosis	Surgeons	Semi-structured interviews, online survey and Nominal Group process
Crowley *et al* (2012)	ACE-inhibitors	Doctors, patient representative, pharmaceutical representative, insurer representative	Moderated group discussions: expert input and ranking
Dear *et al* (2012)	Colorectal cancer	Consumers, health care professionals, researchers and funding agencies	Evidence search then consensus meeting
Deutschman *et al* (2012)	Critical care research	Professional bodies and experts	Steering committee and working groups conference calls then meeting with breakout sessions
Doorenbos *et al* (2008)	Oncology nursing	Oncology nurses	Online survey
Drennan *et al* (2010)	Nursing	Nurses	Delphi survey plus discussion workshop
Dzidowska *et al* (2010)	Psycho-oncology	Researchers and clinicians	Focus groups used to develop survey; survey to rank
Eapen *et al* (2013)	Cardiovascular disease	Clinical, policy, and patient perspectives.	**Stakeholder consensus:** stakeholder identification, environmental scan to identify gaps and prioritised by voting
Elena *et al* (2013)	Cancer outcomes	Researchers	Expert workshop
Esmail *et al* (2013)	Cancer genomics	Patient advocates, payers, clinicians, policymakers, regulators, industry	Landscape analysis (literature review and expert consultation) and modified Delphi process with online voting and stakeholder meeting: **qualitative** content-analysis approach to investigate the themes of the meeting discussion.
Fee *et al* (2011)	Patient safety	Researchers, medical directors, department chairs, hospital administrators, policy-makers.	Multistep nominal group technique at consensus conference
Fletcher-Johnston *et al* (2011)	Paediatric to adult transition in health care	Clinicians and academics	Delphi method
Foltin *et al* (2010)	Paediatric prehospital research	Clinicians and researchers	National consensus project: ranking, scoring against modified Hanlon method and moderated

			discussion to reach consensus.
Foster *et al* (2009)	Musculoskeletal problems	Researchers and patients	**Qualitative approach** and nominal group technique
Gagliardi *et al* (2008)	Colorectal cancer health services research	Researchers, clinicians, managers, policy-makers	**Mixed methodology approach:** gaps identified by systematic review; ranking of gaps by survey; workshop to confirm top-ranked gaps; small group deliberation, full group consensus.
Govindarajan *et al* (2010)	Emergency care networks	Experts: clinical, academic, policy and patients	Consensus conference with breakout sessions
Griffiths *et al* (2002)	Mental health	Researchers, funders, clinicians, mental health consumer and carer advocates.	Questionnaire
Halkett *et al* (2012)	Radiation therapy patient care	Oncologists	Delphi-like study
Harrison *et al* (2006)	Gynae oncology	Oncologists	Mailed questionnaire
Haukoos *et al* (2009)	Sexual health in emergency departments	Academic and clinical experts	Electronic survey and nominal group technique at consensus conference
Henzel *et al* (2011)	Pressure ulcer management	Clinicians and researchers	Consensus panel and survey
Holgate (2007)	Respiratory research	Researchers and funders	Interactive consensus workshop
Howell *et al* (2012)	Anaesthesia	Anaesthetists and lay representatives	Questionnaire and expert panel
Issel *et al* (2012)	Public health nursing	Multidisciplinary experts	**Mixed methodology:** Consensus conference, round table with further consensus then ranking survey
Jagsi *et al* (2012)	Radiation oncology	Clinicians, trainees, researchers, government, industry and payers	**Mixed methodology:** Structured interviews, focus groups, survey questionnaire and three round Delphi process
Jensen *et al* (2011)	Emergency medical services	Researchers, educators, providers, management, regulators, medical directors	**Mixed methodology study:** qualitative baseline interviews, facilitated roundtable discussion, Delphi consensus survey.
Kaji *et al* (2010)	Medical-surgical emergency research	Members of the NIH Task Force and academic leaders in emergency care	Expert roundtable: Domain experts identified research priorities. Lists were circulated among participants and revised to reach a consensus.
Keirns (2009)		Charities, patient organisations, doctors, scientists.	**Ethnographic study:** stakeholder interviews, participations in community forums, observation of coalitions.
Kenward *et al* (2007)		A&E specialists	Modified Delphi method
Kim *et al* (2002)		Clinical and academic nurses	2 round Delphi survey and expert panel workshop
Koutroumanos et al (2013)	Birdshot uveitis	Patients, carers, health professionals	**Qualitative study:** surveys, questionnaires, interactive poster boards and semi-structured

			interviews.
Kraiss *et al* (2013)	Vascular research	Clinicians, researchers	Survey; meeting using nominal group technique
Kramer (2005)	Risk management	Government, academia, industry, healthcare organisations, consumers.	Workshop with modified nominal group process
Krishnan *et al* (2013)	COPD	Patient advocacy groups, health plans, professional organisations representing clinicians, quality improvement organisations, industry, research organisations, government agencies	Modified Delphi approach; workshop using the analytic hierarchy process.
Krumholz *et al* (2005)	Cardiovascular disease	Outcomes researchers	Working group
Lenaway *et al* (2006)	Public health systems research	Public health practitioners and researchers	"Consensus based": Brainstorm session, meeting with nominal group process, dissemination and feedback.
Lewin *et al* (2011)	Community care research	Academics/ researchers, consumers, providers, funders, policy-makers, professional organisations	4 round Delphi process
Li *et al* (2012)	Primary open-angle glaucoma	Members of the American Glaucoma Society	2 round Delphi, identify gaps with systematic reviews in 45 areas then combine results
Loeb *et al* (2001)	Infections in older adults	Researchers, healthcare professionals, providers, government, industry	Workshop: mixed breakout sessions to develop research questions. These were ranked, discussed and re-ranked.
Lowry *et al* (2012)	Cancer care	Clinicians	**Mixed methodology:** structured interviews
Malcolm *et al* (2008)	Children's hospice care	Families, hospice staff and volunteers, professionals associated with the hospice	**Qualitative:** semi-structured interviews, focus groups, telephone interviews. Interpretive analysis.
Mamaril *et al* (2009)	Perianaesthesia nursing	Perianaesthesia nursing experts	3 round Delphi method: electronic surveys
Mcilfatrick *et al* (2003)	Cancer nursing	Nurses	Delphi method
Miller *et al* (2008)	Paediatric emergency care	Members of the Pediatric Emergency Care Applied Research Network	**Mixed methodology:** Nominal Group Process and Hanlon Process of Prioritization
Mindell *et al* (2006)	Paediatric insomnia	Researchers, clinicians	Conference: Plenary presentations and discussions then working sessions.
National Hospice and Palliative Care Organization (2004)	Hospice and palliative care	Researchers	Research conclave: Expert presentations, discussions and deliberation

Nierse *et al* (2013)	Neuromuscular disease	Patients and professionals	**Dialogue model:** Interviews, focus groups, expert meetings, dialogue meeting.
NINDS ICH, W. P. (2005)	Spontaneous intracerebral hemorrhage	Research and clinical experts	Workshop
O'Dwyer *et al* (2007)	Colorectal cancer	Community and academic oncologists, patient advocacy groups, pharmaceutical industry' governmental agencies	Multidisciplinary panel
Owens *et al* (2006)	Paediatric sleep medicine	Researchers, clinicians, research funders, patient education and advocacy groups, industry.	Conference: plenary presentations, multiple discussion panels and work groups. Working groups reported back in large group session.
Paul *et al* (2011)	Psychosocial research in haematological cancers	Patients, carers, clinicians, researchers, social workers, psychologists, patient advocates.	Two-step value-weighting approach: modified Delphi approach, then resource allocation (value-weighting) survey.
Proser *et al* (2007)	Community Health Centre research	Researchers, health policy experts, CHC representatives, primary care associations, government agencies, foundations, journals.	Meeting to produce consensus-driven research agenda: background papers summarising knowledge gaps and feasibility of research; papers discussed then breakout sessions.
Ramelet *et al* (2012)	PICU nursing	Nurses	Delphi method
Ramos-Gomez *et al* (2005)	Oral health	Health care providers, educators, academicians, researchers and policy experts	Workshop: MEDLINE search led to five areas for review. Plenary session then small group workshops using nominal group technique.
Robbins *et al* (2010)	Pulmonary vascular disease	Multidisciplinary experts	Workshop of multidisciplinary experts
Rondinone *et al* (2010)	Occupational safety and health	Occupational medicine, health and prevention professionals	Survey based on previous Delphi round
Ropka *et al* (2002)	Oncology nursing	Nurses, researchers	Cross-sectional mailed survey
Rosen *et al* (2006)	PDE-5 inhibitors and sexual behavior	Researchers, government representatives, scientific and regulatory agencies, pharmaceutical industry	Conference with working groups
Rudolph *et al* (2009)	Paediatrics in secondary care	Paediatricians	Delphi study
Saldanha *et al* (2013)	Gestational diabetes mellitus	Researchers, clinicians	Systematic review, survey, interactive meeting and Delphi process
Sawin *et al* (2012)	Paediatric nursing	Paediatric nurses	Modified Delphi technique
Schroen *et al* (2008)	Breast cancer surgery	Surgeons	Mailed survey
Shah *et al* (2012)	Heart transplantation research	Researchers	Working Group of experts

Sheikh *et al* (2008)	Selected topics in respiratory research	Clinical researchers, academics	eDelphi exercise
Sinaii (2010)	Healthcare-associated infections	Epidemiologists	Electronic and paper survey
Smith (2010)	Occupational health and safety	Academics, patient representatives, government	Modified Delphi technique: expert presentations, small group work to identify priorities, group discussion to list broad areas.
Sparano *et al* (2010)	Breast cancer therapy	Researchers	Expert panel consensus
Tong *et al* (2008)	Chronic kidney disease	Patients	**Qualitative approach:** focus groups
Vella *et al* (2000)	Critical care	Doctors and nurses	Survey for topics, nominal group technique in meeting, survey to check findings.
Ward *et al* (2013)	Childhood obesity	Academics, funders of research, government and policy-makers, healthcare professionals	Multidisciplinary expert meeting and ranking survey
Whitehead *et al* (2004)	Fecal and urinary incontinence	Multidisciplinary with lay advocates for patients	Consensus conference; multidisciplinary experts submitted top research priorities. Discussed at conference then experts asked to revise initial priorities. Steering committee integrated initial and final priorities.
Willett *et al* (2010)	Orthopaedic trauma	Orthopaedic surgeons and nurses	Delphi approach with literature search of top 10 to assess knowledge and research projects.
Wright *et al* (2006)	Cancer research	Patients and carers	**Participatory research:** focus groups in 3 phase process. Pilot phase, then 2 phases of focus groups with different sampling techniques.
Wright *et al* (2012)	Health care infections	Infection Control professionals, epidemiologists	National survey around topics identified by previous Delphi exercise
Wu *et al* (2003)	Bone metastases	Radiation oncologists	One day workshop: Initial survey to identify research areas. Workshop with plenary session followed by subgroups to deliberate and list prioritised trials
Yano *et al* (2006)	Healthcare of women veterans	Researchers	Systematic review then conference: plenary sessions then 5 moderated workgroups.
Yano *et al* (2011)	Healthcare of women veterans	Researchers, policy and practice officers, Congressional staffers, patient representatives	Conference: plenary and breakout sessions
Yassi *et al* (2005)	Infection control in SARS	Health care workers, support staff, hospital managers	Literature review and focus groups to identify gaps, key areas identified by consensus
Yin *et al* (2000)	Nursing research	Nurses	**Participatory:** Conference with ranking exercise. Self-described as participatory, from text not traditionally participatory.

Table 30 Summary of non-ENT, Hearing and Balance care RPSEs

Appendix 4: A comparison of the research priorities from previous ENT, Hearing and Balance care RPSEs

Netherlands ENT Society Exercise: Otology	JLA Tinnitus PSP
Primary research questions 1. What is the value of an MRI scan to prevent unnecessary 'second look' operations after previous cholesteatoma surgery? How often must this MRI be performed after surgery? Is this cost effective? 2. What are the factors that determine the effectiveness of speech understanding in background noise with: bilateral hearing aids, hearing aid and cochlear implant, or bilateral cochlear implants? 3. What is the most effective therapy for granulomatous otitis externa? *Guidelines* 1. Guidelines for the diagnosis of patients with a cerebellopontine angle mass. 2. Guidelines on aetiology, diagnostics and treatment of tinnitus. 3. Guidelines on the indication for auditory implants. *Other* 1. Set up a registry for ear surgery outcomes with the aims of epidemiological analysis, establishment of outcomes standards and quality improvement. 2. Combine the priorities of this ENT scientific agenda with those of the national hearing screening program (NPG) of the National Hearing Foundation following their development. (van Benthem *et al* 2013)	What management strategies are more effective than a usual model of audiological care in improving outcomes for people with tinnitus? Is Cognitive Behaviour Therapy (CBT), delivered by audiology professionals, effective for people with tinnitus? Here comparisons might be with usual audiological care or CBT delivered by a psychologist. What management strategies are more effective for improving tinnitus-related insomnia than a usual model of care? Do any of the various available complementary therapies provide improved outcome for people with tinnitus compared with a usual model of care? What type of digital hearing aid or amplification strategy provides the most effective tinnitus relief? What is the optimal set of guidelines for assessing children with tinnitus? How can tinnitus be effectively managed in people who are Deaf or who have a profound hearing loss? Are there different types of tinnitus and can they be explained by different mechanisms in the ear or brain? What is the link between tinnitus and hyperacusis (over-sensitivity to sounds)? Which medications have proven to be effective in tinnitus management compared with placebo? (Hall *et al* 2013)

Table 31 A comparison of tinnitus priorities from previous exercises

Netherlands ENT Society Exercise: Balance	JLA Balance PSP
Primary research questions Research into the effectiveness of different interventions in the treatment of specific patient groups with dizziness (such as Ménière patients): intra-tympanic treatment with medications such as corticosteroids or gentamicin, behavioural therapy, surgical interventions, physiotherapy. Development and evaluation of diagnostics of the different components of the balance system. *Guidelines* Multidisciplinary diagnostic guidelines for patients with dizziness. *Other* No suggestions	What is the optimal process for GP education and training for improved diagnosis and management of balance disorders? What is the best way of training health professionals in the management of balance disorders? What is the most effective treatment for Vestibular migraine? What are the best interventions to improve balance/minimise symptoms in daily activities such as supermarkets, escalators etc? Is any specific surgical intervention effective in Meniere's disease and what procedure is best? Are there any effective interventions for the ear pressure symptoms in Meniere's disease? What is the optimum pharmacological strategy for the management of patients with Meniere's disease? In particular, what are the effects of betahistine (including long term effects)? Is it helpful in preventing the severity, frequency and progression of attacks of Meniere's disease to adopt a specific diet, or restrict salt, caffeine or fluid intake? Are the home-based exercises given to patients with balance disorders effective? Are stress management techniques helpful in patients with balance disorders?
(van Benthem *et al* 2013)	(JLA Balance PSP 2011)

Table 32 A comparison of balance priorities from previous exercises

Appendix 5: The GENERATE survey

The GENERATE Survey
Working together to develop research that matters in ENT, Hearing and Balance

This is an exciting opportunity to have your say in what you think is important for future research for ENT, Hearing and Balance care in the UK. We want to know from your experience what you think are the most important areas for research and why you consider them important. Your ideas will be brought together and shared with our partners in order to prioritise, develop and find funding for the top projects. This project is a partnership of healthcare professionals, patients and their carers, researchers, charities and people who design and deliver ENT, Hearing and Balance healthcare services.

What is ENT?

Ear, Nose and Throat (ENT) conditions affect people of all ages. Most children will have had ear infections by the time they start school, and one in four adults suffers from hay fever. With age we all lose some of our hearing and may suffer from tinnitus or dizziness leading to falls. Healthcare professionals working in the field of ENT also treat cancer of the head and neck, conditions affecting hearing, smell, taste and balance, and problems with the voice, breathing and swallowing.

Why do we need research?

These conditions have a major impact on patients, their families and their quality of life, so it is important that people with ENT, hearing and balance conditions receive the best care. To decide what the best care is more high quality research into the prevention, diagnosis and treatment of these conditions, as well as research into how care is delivered is required.

What is research?

GENERATE covers topics in clinical and health services research in all areas of ENT.

"Clinical research helps us understand how to diagnose, treat, cure or prevent health problems."

NIHR Clinical Research Network. "Do you want a say in clinical research?"
www.crncc.nihr.ac.uk/ppi

Health services research looks at how people get access to health care, and examines patients' experiences of care and their outcomes.

For more information on GENERATE please go to: www.entuk.org/ent_patients/generate

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Examples of topics and questions that <u>other surveys </u>have raised are:

"When is the best time for a person with dementia to move into a care home and how can the standard of care be improved?"

From the Dementia Priority Setting Partnership
www.lindalliance.org/DementiaPSP.asp

How burn survivors get access to mental and social care after they have been in the hospital.

From Broerse *et al* "Involving burn survivors in agenda setting on burn research: An added value?"
www.elsevier.com/locate/burns
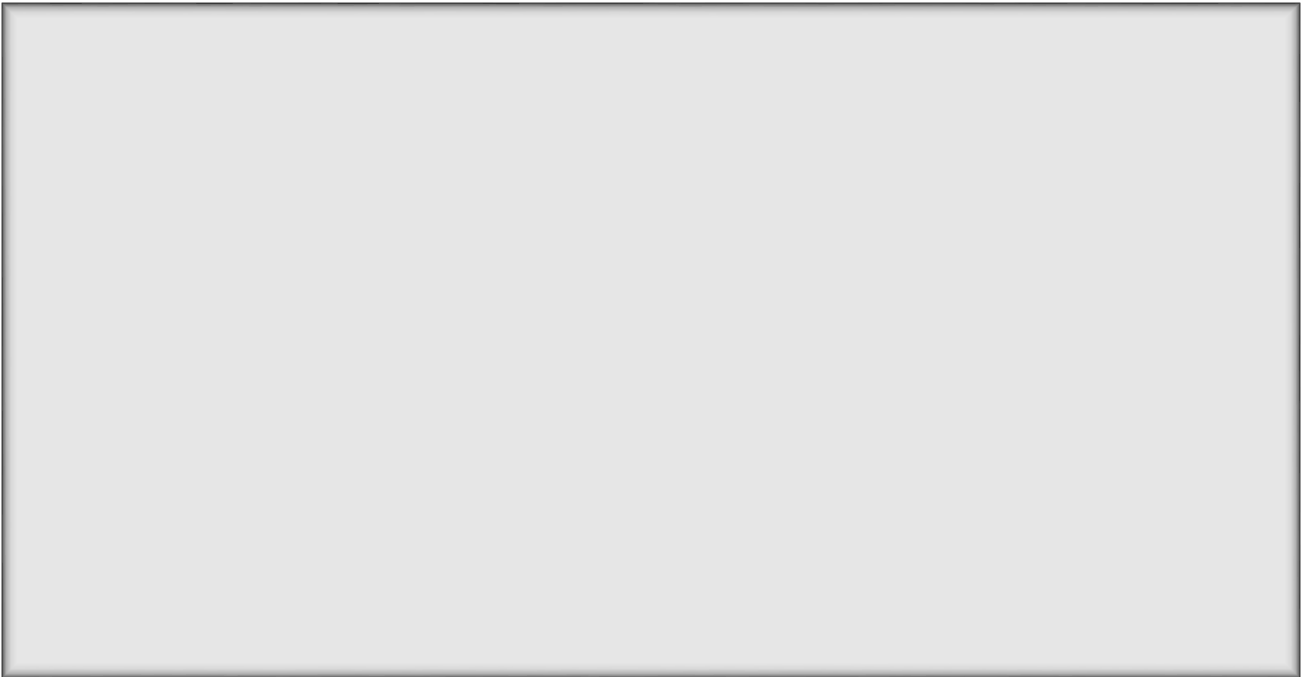
The role of diet to prevent the progression of chronic kidney disease.

From Tong *et al* "patients' priorities for health research: focus group study of patients with chronic kidney disease"
www.ndt.oxfordjournals.org

HOW TO TAKE PART: <u>Based on your experience</u>, please answer the following two questions:

What do you think are the most important topics, questions or issues that should be answered by research for ENT, Hearing and Balance care?

Why do you think these are important?

This survey is anonymous. We would like some information about you so that we can make sure that we have a wide range of opinions, but the following questions are **optional**. You do not have to answer them.

What group do you belong to? Please choose the one that best describes you:
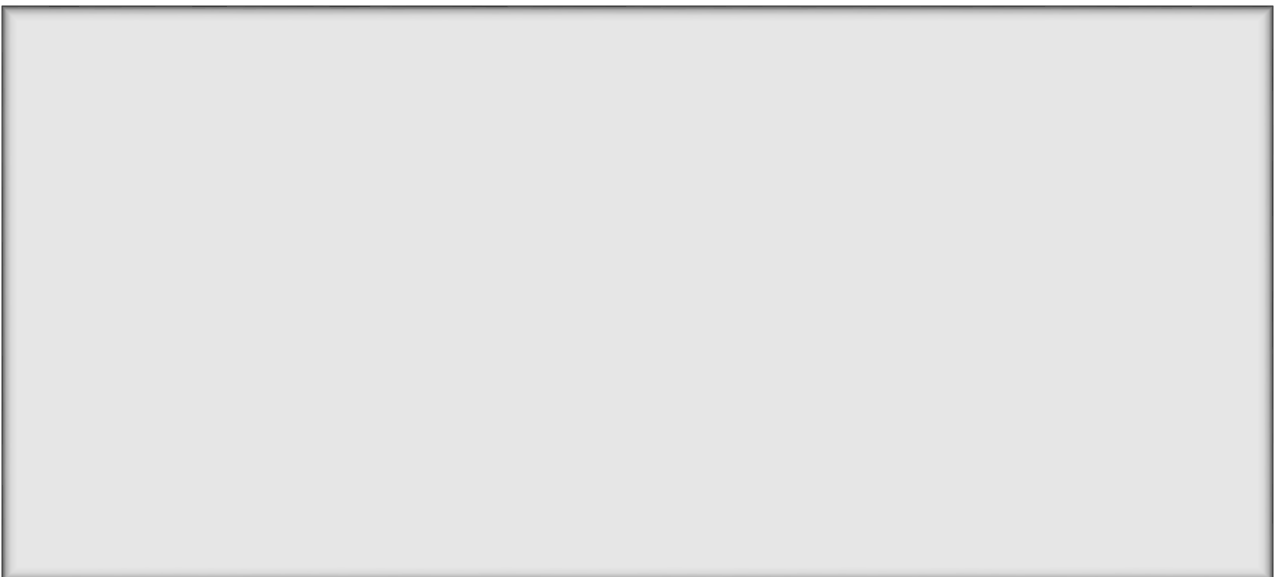
Patient or member of the public:

Patient…………………………………☐	Other………………………………… ☐
Patient Carer…………………………☐	*If 'other', please specify:_____*

Clinician:

Anaesthetist……………………………☐	ENT Junior Doctor………………… ☐
Audiologist……………………………☐	GP………………………………………. ☐
Audiovestibular Consultant……. ☐	GP w/special interest in ENT… ☐
Audiovestibular Associate	Nurse………………………………….. ☐
Specialst………………………………. ☐	Speech & Language Therapist… ☐
Audiovestibular Junior Doctor….☐	Other………………………………… ☐
ENT Consultant……………………. ☐	*If 'other', please specify:*
ENT Associate Specialist………… ☐	_____

Clinical Researcher:

Professor……………………………….. ☐	Academic Clinical Lecturer…….. ☐
Senior Lecturer…………………………. ☐	Academic Clinical Fellow……….. ☐

Please state your specialty: _____

Academic:

Professor……………………………….. ☐	Doctoral researcher……………. ☐
Post-doctoral researcher…………. ☐	

Please state your specialty: _____

Clinical Commissioner:

CCG member………………………………☐	Other……………………………….. ☐

If you have selected 'other', please specify:_____

Service Manager:

NHS Manager……………………………. ☐	Other……………………………………..☐

If you have selected 'other', please specify:_____

Which category includes your age?

17 or younger…..	☐	50 – 59…………	☐
18 – 20…………….	☐	60 – 69…………	☐
21 – 29…………….	☐	70 – 79…………	☐
30 – 39…………….	☐	80 or older……	☐
40 – 49…………….	☐		

What is your gender?

Male…☐   Female…☐

Where are you from in the UK?

England…☐   Northern Ireland…☐   Scotland…☐   Wales… ☐

What is your ethnic group? Please choose one option that best describes your ethnic group or background:

White……………………………………………	☐
Mixed/multiple ethnic groups………………..	☐
Asian/Asian British……………………………….	☐
Black/African/Caribbean/Black British……	☐
Other ethnic group………………………………	☐

*If you have selected 'other', please specify:*_____

☺ *Thank you for completing this survey!* ☺

Please hand this back in or send it to:
Natalie Bohm
ENT UK at the Royal College of Surgeons
35-43 Lincoln's Inn Fields
London, UK
WC2A 3PE

The survey may be accessed online at:
https://www.surveymonkey.com/s/ENTUK_GENERATE

Appendix 6: Details of survey dissemination

The survey was open from 28 April 2014 to 30 September 2014. Of the 467 stakeholder organisations identified for the survey, 383 organisations were contacted and asked to disseminate information and the electronic link to the survey. The information was in the form of press releases or short articles to be distributed through membership email lists, newsletters, websites and social media. 84 organisations were not contacted either because they no longer existed or due to a lack of up to date contact details.

The table below shows which of the stakeholder groups disseminated the survey, which did not respond to the request for dissemination and which declined to disseminate it. Of the organisations that did not respond to the request, it is unclear whether they did forward information on the survey to their memberships or contacts.

Stakeholder Group	Number of organisations			
	Publicised survey	Did not respond	Refused survey	Total contacted
Patient/Public	18	55	4	77
Healthcare Professionals	30	31	4	65
Allied sectors	1	3	0	4
Managers/Commissioners	10	227	0	237
Total numbers	59	316	8	383

Table 33 Stakeholder groups and survey dissemination

30 of the 59 organisations that publicised the survey were Healthcare Professional organisations, making this the largest stakeholder category to disseminate it. The second largest stakeholder category was the Patient and Public category, where 18 organisations sent information to their memberships about the survey. One organisation from the Allied Sectors, the British Association of Teachers of the Deaf, circulated the survey to its membership. From the Managers and Commissioners stakeholder category, the largest response came from the Welsh Health Boards, where six of the seven Boards publicised the survey. Two English Clinical Commissioning Groups and one Northern Ireland Local

Commissioning Group disseminated the survey; none of the Scottish Health Boards contacted responded. Also in this stakeholder group, the NHS Foundation Trust Network, an organisation for NHS managers circulated the survey link to its members.

The largest group of non-responders to the request to publicise the survey were from the Managers and Commissioners stakeholder group. The majority of organisations that did not respond were English Clinical Commissioning Groups (208 out of 227 non-responders). All non-responders had repeat email requests and letters sent to them, where contact details were available.

Eight organisations declined to publicise the survey. The reasons given for this were:

- that there was an organisational policy against promoting surveys;

- the organisations did not feel that the survey was relevant to its membership;

- the organisations felt that their membership had received the survey through other channels due to an overlap of membership with another organisation.

In addition to contacting stakeholder organisations, the clinical directors or clinical leads of UK hospital ENT departments from the ENT-UK database were contacted by email and asked to circulate the survey to their clinicians and managers. 63 clinical leads/directors were identified and contacted. They were also asked if they would distribute paper copies of the survey to patients in their Outpatient Departments. Four hospitals agreed to distribute paper copies to patients (three hospitals in England and one in Northern Ireland).

Finally, in order to promote awareness of the survey and increase the response rate, presentations on the GENERATE project with the survey link were given at four national professional conferences and two local (London) academic meetings. Paper copies of the surveys were distributed at each of these meetings. Paper copy surveys were also made available to patients and members of the public at two London events: the National Institute

of Health Research International Clinical Trials Day, and the University College London

Hospital Research Open Day.

Due to the method of distributing the survey through external organisations with private

membership lists, as well as distribution through social media and public events, data on the

number of recipients of the survey are not available.

Appendix 7: Focus group topic guide

What do you understand by ENT, Hearing and Balance care?

Prompts:

Hearing; tinnitus; dizziness; vertigo. Cancer of the head and neck; head and neck infections; colds; ear infections. Conditions affecting hearing, smell, taste and balance. Problems with the voice, breathing and swallowing. Ages treated: children, adults, the elderly.

Professionals involved in treatment: ENT, audiology, audiovestibular medicine, SLT, GPs, etc.

Place of treatment: community, home, primary care, hospital, etc.

What do you understand by research?

Prompts:

Clinical research, understand/investigate/test how to diagnose, treat, cure or prevent health problems.

Health services research, access to care, patient pathway, information, patient experiences, patient outcomes.

Based on your experience what are the most important topics, issues or questions that would benefit from research in ENT, Hearing and Balance care?

Prompts: To be developed from emerging framework from survey.

Why do you think these are important?

Prompts: To be developed from emerging framework from survey.

Which of the topics discussed today do you think are the most important for further research?

Why are they the most important topics?

Appendix 8: Focus group participant information

Setting the research agenda for ENT, Hearing and Balance care

Thank you for agreeing to take part in this discussion that is a key part of the joint effort of ENT UK, the British Society of Audiology and the British Academy of Audiology to develop a research agenda for ENT, Hearing and Balance care in the UK. We are looking at areas of clinical and health services research, as we recognise that high quality, focused research in these areas is needed to ensure the sustained delivery of high value care in our field.

The purpose of the focus group is to discuss what areas of ENT, Hearing and Balance care are most in need of further research. This can be either clinical or health services research. We also want to understand why you think these areas are important. We are interested in your opinions based on your unique perspective, experience and expertise. As part of the conversation we will ask you to share some of your experiences of commissioning or providing healthcare and where you feel evidence is needed to inform your decisions. We will also explore whether the areas under discussion fall into certain themes. We are not looking for "right answers," rather we want to hear the discussion that comes from different points of view.

Figure 1: The agenda-setting process
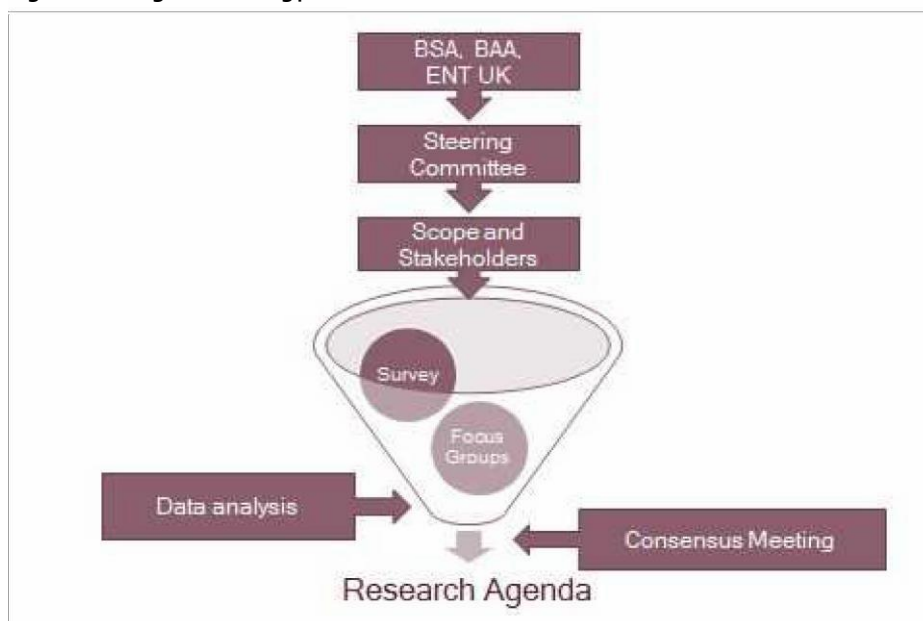
We have already gathered some ideas through a survey. We are also using focus groups to bring together key people in order to explore their opinions in greater depth. All the data gathered will be qualitatively analysed using a framework analysis (Ritchie, Spencer *et al* 2003).

This analysis will form the basis for discussion at a consensus meeting in 2015 to establish the proposed agenda (see Figure 1 overleaf for a diagrammatic overview of the process). Participants at this meeting will be representative of the stakeholders from whom we have gathered our data and will be agreed by our independent Steering Committee. The results of this will be available to all have participated in the agenda-setting process.

The discussion will be informal and will follow a focus group format with everyone being encouraged to express their views. It will be moderated by Natalie Bohm, Clinical Academic Lecturer ENT Surgery UCL. The discussion will be audio-recorded for transcription. All responses will be anonymous in transcription, and the original recording will be destroyed. Only the research team and focus group members will know your identity. As this is considered to be service improvement, formal research ethical approval has been deemed unnecessary; nevertheless all data will be kept secure in UCL computers. At the end of the session you will be asked to fill in a brief feedback form so that we can improve our future consultations.

Thank you again for your support. We look forward to seeing you on **Thursday 11th December, 15.00 – 16.30, evidENT unit, Royal National Throat, Nose and Ear Hospital, 330 Gray's Inn Road, King's Cross, London, WC1X 8DA**.

Please contact Project Administrator **Marianne Simmonds** on **0203 108 9244** or at **m.simmonds@ucl.ac.uk** if you have any questions.

References

Ritchie J, Spencer L and O'Connor W. (2003). Carrying out qualitative analysis. In Ritchie J. and Lewis J., eds. *Qualitative Research Practice* London: Sage Publications Ltd. Ch 9.

Royal National Throat, Nose and Ear Hospital/Ear Institute
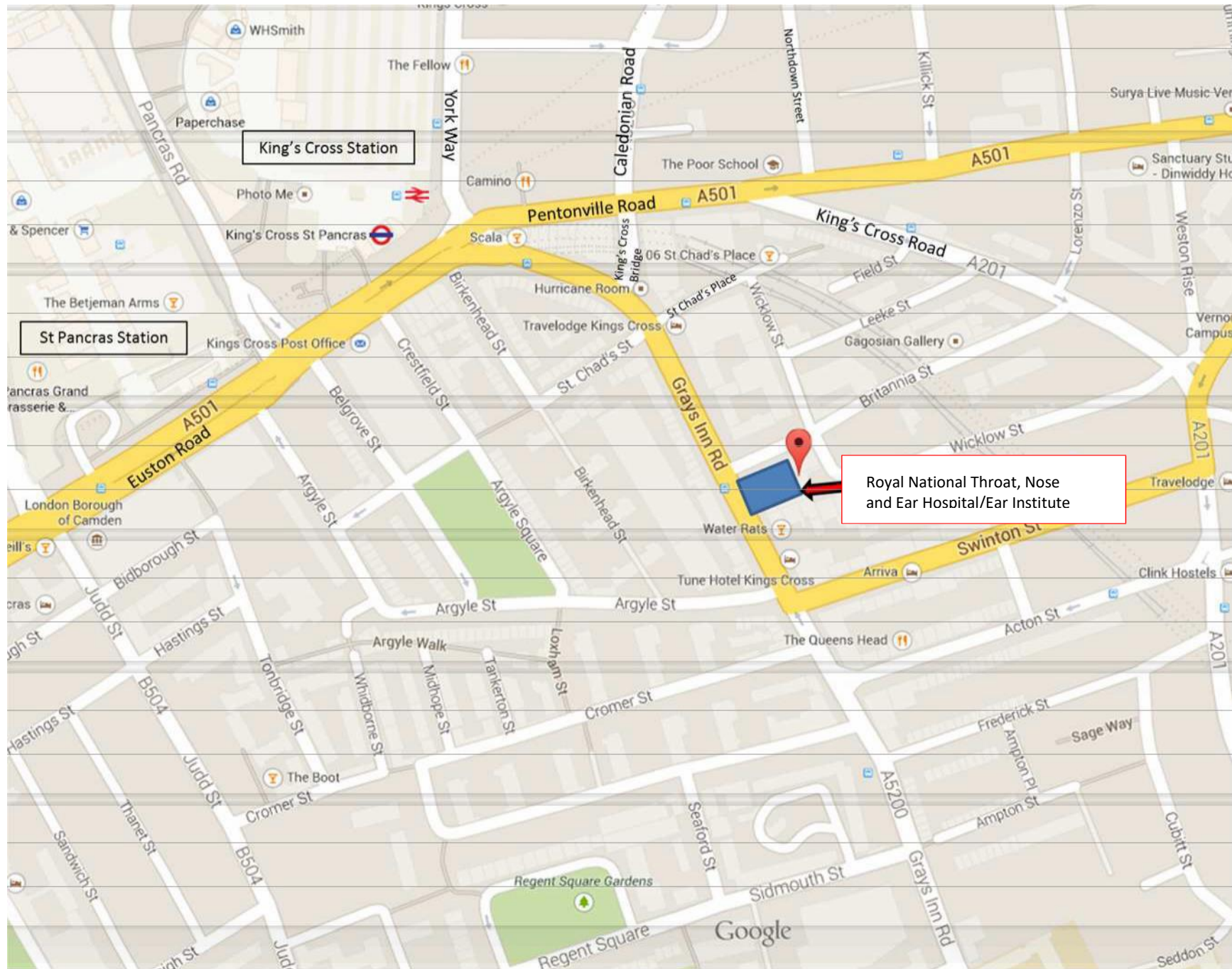
Entrance of the Royal National Throat, Nose and Ear Hospital if you are walking along Gray's Inn Road away from King's Cross/St Pancras stations
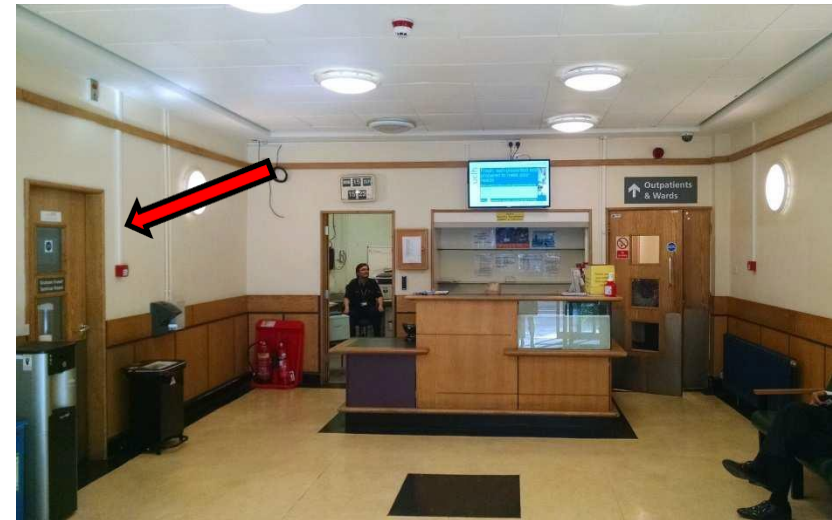

Entrance of the Royal National Throat, Nose and Ear Hospital if you are walking along Gray's Inn Road towards King's Cross/St Pancras stations

Entrance of the Royal National Throat, Nose and Ear Hospital from across the road

Take the door on the left, then the door marked 'evidENT', go to the very top of the stairs, turn right and ring the doorbell.

About the GENERATE initiative

Introduction
The GENERATE project is an exciting opportunity for healthcare professionals, patients, carers, commissioners, service managers and members of the public to have their say in what they think is important for future research for ENT, hearing and balance care in the UK.

Who is behind GENERATE?
GENERATE is a joint research project, being run by ENT UK, the professional body of Ear, Nose and Throat surgeons, in conjunction with the British Academy of Audiology (the largest association of professionals in hearing and balance in the UK) and the British Society of Audiology (the largest Audiology society in Europe). GENERATE is wholly non-commercial.

Why?
Hayfever, ear infections, hearing loss, dizziness - most people suffer from an Ear, Nose and Throat (ENT) condition at some stage in their life.

Research will help to prevent and cure ENT conditions. These problems can have a major impact on patients, their families and their quality of life. It is important that people with ENT, hearing and balance conditions receive the best care. Furthermore, as healthcare budgets are tightened, we need to ensure that treatment and care are as effective, and therefore efficient, as possible. To determine the best care for patients with these conditions, more high quality research into the prevention, diagnosis and treatment of these conditions, as well as research into how care is delivered, is required.

How?
The GENERATE survey took place over summer 2014, collecting almost 700 responses. These give the views of patients and their carers, healthcare professionals, researchers, charities and people who design and deliver healthcare services and will help to decide the most important areas for research in ENT, Hearing and Balance care.

You have been invited to take part in a focus group (discussion group), so that we can explore the views of commissioners and service managers in more detail.

What next?
The ideas collected from the surveys and focus groups will be brought together and shared with our partners in order to prioritise, develop and find funding for the top projects. The project will develop a joint national programme for research.

All the information – including your responses - will be anonymous.

Do I have to take part?
Not at all! Participation is voluntary. It would be helpful if you could let us know if you will not be attending the focus group, so that we can amend our catering order!

Questions?
Please contact Marianne Simmonds, Project Administrator on 0203 108 9244 or at
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Appendix 9: Long list of research ideas

Adult hearing loss and tinnitus
Understanding Health and Illness
Incidence and prevalence:
What is the burden of disease in hearing loss and acoustic neuroma? What is the prevalence of tinnitus in hypothyroidism?
Investigating the role of the following risk factors in causing or worsening conditions:
•Diet/stress and hearing loss/tinnitus
•Smoking and hearing loss
•Noise and hearing loss (personal listening devices; workplace noise hazards; individual risk factors)
•Hormones/infections/viruses and tinnitus
•Mobile phones and acoustic neuromas
Aetiology: How do we determine the underlying cause or causes to develop preventative measures or treatments for hearing loss, tinnitus, acoustic neuroma and hyperacusis?
Links to other conditions:
What is the link between acute hearing loss and thromboembolic disease/stroke?
The link between tinnitus and hypothyroidism: does treatment of the latter improve the former? What is the link between tinnitus and hyperacusis?
What is the effect of fibromyalgia on hearing?
Is there a link between acoustic neuroma and neurological conditions and other tumours such as meningioma? Is there a link between hearing loss and snoring?
Can hearing be affected by a laryngectomy?
Pathogenesis:
Understanding the mechanisms behind auditory processing: can improving cognition improve hearing? What is the pathogenesis of acoustic neuroma?
What is the mechanism of tinnitus?
Genetics and heredity: Are hearing loss, tinnitus or acoustic neuroma hereditary?
Understanding how conditions progress:
Long term follow up of people who have had ototoxic drugs to determine long---term outcomes.
Understanding the natural history of retraction of the tympanic membrane and development of cholesteatoma; what is the natural history of cholesteatoma in adults?
What is the natural history of acoustic neuroma: what is the relationship between loss of balance and the speed at which an acoustic neuroma develops.
Understanding plasticity and hearing, especially in unilateral hearing loss.

Indirect healthcare costs:

What are the financial implications to the government of noise---induced hearing loss in the military? What is the socioeconomic impact of single---sided deafness?

What is the financial impact of hearing loss on families?

Quality of life:

Understanding how having a "hidden" condition (hearing loss including single---sided deafness; tinnitus) increases frustration and suffering due to the impact on social interaction, family interaction, interaction with healthcare professionals, enjoyment of life and external perceptions of intelligence.

What is the general quality of life for patients with tinnitus; cholesteatoma or facial nerve damage?

Social impact:

What is the impact of unaddressed hearing loss, single---sided deafness and tinnitus on social isolation and withdrawal? Would screening/fitting hearing aids at an earlier stage reduce social isolation?

What is the social aspect of hearing aid use, such as the perception of stigma?

Relationships: What is the effect of hearing loss on relationships?

Impact on work and employment:

What is the impact of hearing loss/distorted hearing/tinnitus on current employment, future opportunities or seeking employment?

What is the effect of negative information in audiology about decreased sound tolerance (tinnitus/hyperacusis) on patient and family wellbeing?

How do tinnitus, hearing problems, and single---sided deafness disrupt a "normal," healthy life and being able to engage in "normal" activities? What is the degree of fatigue caused by acoustic neuroma/hearing dysfunction and how does this impact on daily living?

What are the levels of distress/daily suffering caused by tinnitus, hyperacusis, and acoustic neuroma to different individuals, and how do we assess this?

Prevention of Ill Health, Maintenance of Good Health

Primary prevention:

Prevention of noise---induced hearing loss and tinnitus:

•Is there any benefit of regulating environmental noise including traffic/cinemas/coffee machines on noise--- induced hearing loss?

•What is the impact of campaigns about use of personal music devices/headphones/mobile phones on the development of noise---induced hearing loss and tinnitus?

Ototoxicity:

•What is the impact of increasing awareness and avoidance of ototoxic agents?

•Can we develop a simple test to risk assess ototoxicity for eardrops?

•Assessment of the long---term risks of repeated eardrop use with perforation. Is there a role for diet in preventing hearing loss?

Secondary prevention: Would a hearing screening programme for adults newly diagnosed with dementia be cost effective?

Tertiary prevention:

Hearing rehabilitation:

•How can you optimise residual hearing through hearing rehabilitation? Do hearing training programs optimise the effectiveness and uptake of hearing aids?

•What is the clinical and cost---effectiveness of wider hearing rehabilitation on hearing loss?

•What strategies provide effective rehabilitation in severe/profound hearing loss?

•What should the key outcomes be for adult auditory rehabilitation? What are the long---term outcomes of adult auditory rehabilitation? What is the impact of hearing rehabilitation on quality of life/social interaction/cognition?

•What factors improve long-term adherence to therapy in adult auditory rehabilitation?

•What are effective rehabilitation strategies for single-sided deafness?

Tinnitus rehabilitation:

•What is the role of, and support for, lifestyle changes in tinnitus rehabilitation?

•What is the impact of tinnitus rehabilitation on quality of life? What is the effectiveness and role of tinnitus rehabilitation in managing symptoms?

•Is there a role for mindfulness in tinnitus rehabilitation?

Promoting self-help:

•How can technology be used to promote independence: what is the accessibility of assistive technology for hearing and the role of self-adjustment of hearing aids?

Clinical Effectiveness

Diagnostic testing and assessment:

How do we improve assessment of residual hearing in order to optimise rehabilitation:

•Can suprathreshold aspects of hearing be reliably measured using auditory evoked potentials (AEPs)? What are the best approaches and how do they compare to psychophysical approaches? What is the role of speech in noise testing in adult aural rehabilitation?

•Is there another diagnostic tool to supersede the conventional audiogram that can improve our understanding of the individuals hearing loss?

Develop objective audiological testing:

•Can objective parameters reliably be used as an alternative to subjective judgement of response presence? Do speech in noise tests better reflect real life patient difficulties as opposed to objective tests?

•Can the use of objective measures speed up clinical measurements such as threshold detection?

•If objective testing is used sequentially how can a desired false positive rate be maintained?

•Multichannel AEP recording: what is the trade---off between the extra time spent in putting on more electrodes and the possible benefits of improved test time or more accurate threshold measurement?

Hearing assessment in adults with learning disabilities:

•What are the optimum electrophysiological tests to assess hearing in adults with learning disabilities?

•What is the best way to test speech discrimination in adults with learning disabilities? How should prescription formulae be adjusted to take into account the perceptual differences to sound experienced by adults with learning disabilities?

Are the levels set by the international standards for Bone Conduction at 2kHz and 4kHz appropriate?

Functional testing for hearing:

•What clinical information may responses to natural stimuli (such as speech or music) indicate that responses to repetitive stimuli such as tone pips do not? How should response to natural stimuli best be measured? Are responses to such stimuli reliable?

Diagnostic radiology: How can diagnostic radiological scanning techniques and interpretation for acoustic neuroma/ cholesteatoma be improved?

Tinnitus:

•Develop diagnostic tools and psychoacoustic tests.

•How can individual severity be assessed?

Diagnostic criteria: How can we improve differential diagnosis/categorisation for tinnitus sub---types? Timeliness and accuracy of diagnosis:

How do we detect/diagnose acoustic neuromas earlier in order to provide a wider range of management options for patients (gamma knife)?

How can we improve accuracy of diagnosis of underlying cause of sensorineural hearing loss to determine treatment?

Improving safety, reducing complications and preventing deterioration:

Acoustic neuroma:

•Following acoustic neuroma removal, how can the loss of facial nerve movement, whether temporary or permanent, be better addressed in respect of loss of taste, problems with sight as well as hearing and general loss of self confidence?

•How can hearing be preserved when treating tumours (acoustic neuroma) with radiation?

•What are the best methods for better methods or equipment to improve hearing and facial nerve outcomes in the surgical management of acoustic neuroma?

What are the optimum intervals for aural microsuction?

Acoustic neuroma: What is the long---term effect of gamma knife treatment? What are the risks and benefits of current acoustic neuroma management?

Comparing interventions:

What are the benefits of digital vs analogue hearing aids; how would patients rate their experience with digital hearing aids compared to analogue?

What are the benefits of aural microsuction over irrigation?

Which treatment option is best to preserve hearing ability/best for overall benefit for people with acoustic neuromas? (Long term benefits of interventions vs watch and wait).

Tinnitus:

•Which medications have proven to be effective in tinnitus management compared with placebo?

•What type of digital hearing aid or amplification strategy provides the most effective tinnitus relief; is there hearing aid that does not amplify tinnitus but screens it out?

•What is the best treatment for tinnitus?

•What is the evidence for auditory implant devices; what are the benefits of cochlear implants vs middle ear implants in patients with severe, not profound, hearing loss?

Otological surgery:

•How can post---operative hearing loss be minimised (acoustic neuroma)?

•What is the best management/ long---term after---care of bone anchored hearing aids to preventing flare---ups from area around the abutment?

Does ear syringing worsen tinnitus?

Timings of interventions:

What effect does the timing of intratympanic steroid injections have on sudden sensorineural hearing loss (optimal time/latest time)?

Hearing aid provision: are there any adverse issues related to delay or non---provision of amplification until a severe hearing loss it reached?

At what stage/size/particular placement of vestibular neuromas do they begin to cause hearing loss and in what ways could early treatment/ intervention prevent this? Would earlier Gamma Knife treatment retain hearing and is the current policy of watch and wait detrimental to patients' wellbeing?

Adopting technology:

Hearing aid and device technology:

•Independent research into use and effectiveness of frequency compression in hearing aids and cochlear dead regions.

•What effect does new hearing aid and implant technology have on communication and listening skills?

•The evidence for adopting newer, more sensitive tools in hearing aid evaluation. Developing and adopting endoscopic middle ear procedures.

Improvement of existing interventions:

Hearing aid and device technology:

•Developing hearing devices to replace natural hearing that work effectively despite the surgical approach used to remove vestibular schwannomas: better cochlear implants and auditory brainstem implants (ABIs).

•Improve the ability of hearing aids to provide natural hearing and hearing in background noise; improve the physical comfort of hearing aids. Hearing aid development trial development: look at how new to market products have been developed, how many individuals have tried them and what research backs up new developments.

•Develop hearing aids that can be integrated into everyday devices (e.g. mobile phones) and that allow the user to have more control over them for adjustment;

•What are the best hearing aids for patients with single sided hearing loss; how do we improve them?

•Optimisation of support equipment for hearing impaired (aids and adaptions); What is the cost effectiveness of supplying radio microphones for hearing aids in the NHS?

Improving outcomes for treatment of vestibular schwannoma, including hearing, facial nerve and balance rehabilitation.

Selecting patients:

Hearing aids:

•Develop the evidence to determine who would benefit from a hearing aid.

•What are the benefits of providing hearing aids for mild---moderate hearing loss? What is the benefit and cost---effectiveness of bilateral cochlear implants in adults? Assessment of current interventions:

Assessing current techniques in Otological procedures: is there a wider role for the use of the laser and would this decrease invasiveness of procedures and complications.

Hearing aids:

•What is the long---term impact of using a hearing aid and the impact of hearing aids on cognition?

•What is the value of hearing therapy in maximising uptake of hearing aids?

•What is the clinical and cost effectiveness of bone conduction hearing devices; What is the effectiveness of BAHAs with magnetic attachment?

Sudden sensorineural hearing loss:

•Do intratympanic steroids work in sudden sensorineural hearing loss?

•Do oral steroids work in sudden sensorineural hearing loss?

Tinnitus:

•Clearer analysis of the main options and their true benefits. Is CBT/psychological therapy, delivered by audiology professionals, effective for people with tinnitus compared with standard audiological care or CBT delivered by a psychologist?
•What is the evidence for tinnitus treatments not widely used in the NHS: acoustic neuromodulation; TMS/rTMS; tinnitus suppression (Soundcure serenade); mindfulness?
Developing novel therapies:
Develop cures or effective therapies for tinnitus/hyperacusis/single---sided deafness. The development of new treatments for cholesteatoma.
Develop treatments for Eustachian tube dysfunction.
Explore the role of emerging communication technology in hearing (e.g. digital communication devices, self-fitting of hearing aids, brain computer interfaces for controlling CIs and hearing aids).
Develop cell---based therapies to allow body to regenerate tissue types in sensorineural hearing loss (consider the Sheffield University Marcelo Rivolta Model); Regeneration of hair cells to restore inner ear hearing.
Restoring lost hearing function through stem cell therapy/gene therapy; restore hair cell and neural function. Nerve regeneration for single---sided hearing loss.
Could fibroblast growth factor instillation replace tympanoplasty for chronic tympanic membrane perforation? Could stem cells be used to repair chronic tympanic membrane perforations?
How can you regenerate facial nerve function?
Using technology to personalise treatment: Develop personalised automated hearing aid fitting; is this cost effective?
Identifying subgroups to customise treatment: Improve differential diagnosis for tinnitus sub-types to develop individualised management of the tinnitus patient.
Outcome measures:
Developing condition specific quality of life outcome measures for gamma knife treatment for acoustic neuroma/cochlear implants (for bilateral implantation).
Use consensus-based approaches to develop outcome measures, especially to determine what domains of impairment are important, for hearing loss and adult auditory rehabilitation.
Is there evidence to supports speech audiometry as a predictor of hearing aid outcome? Develop more functional outcomes measures for hearings aids than PTAs for adults.
Outcome measures to assess benefit from hearing aids for mild-moderate hearing loss.
Designing and Delivering Healthcare
Patient pathways:
Timely diagnosis and referrals:
•What strategies improve delays in referral from primary care to hearing and tinnitus services?
•What is the unmet morbidity from hearing loss due to lack of referral for hearing loss from primary to secondary care?
•What is the impact of delay in referral of tinnitus and hearing loss on patient health, wellbeing and quality of life?
Quality of service:
•What is the impact of improving and developing communication tools for clinics for patients with hearing loss?
•What is the impact of private AQPs on quality of audiology service?
Innovation and service delivery: What is the impact of using technology to develop personalised, automised hearing aid adjustments and fitting?

Fitting pathways to individuals: How do we incorporate diversity of opinion from the deaf community as to which interventions (surgical and non---surgical) should be used in the management of hearing loss?

Follow up:

•What is the benefit of increased follow up for hearing aid users?

•What is the need for ongoing care and follow up of patients following otology procedures? How can the pathway and follow up for people with acoustic neuroma be standardised?

Tinnitus clinics:

•What is the value of a tinnitus clinic in improving quality of life?

•What is the most effective and achievable approach to the management of tinnitus through ENT/audiology collaboration?

Barriers to using services:

Hearing aids and services:

•What are the barriers to people seeking help for their hearing loss and why do they delay in seeking help?

•Investigate and address reasons that hearing aids are underused. What is the value of hearing therapy in maximising uptake of hearing aids? What is the impact on hearing aid design on uptake: the role of aesthetics?

•What strategies improve hearing aid use: providing unilateral or bilateral aids; establishing a baseline audio where optimum usage is most likely to occur; the impact of payment for issue of aids?

Improving access to services:

•How can we improve the equality of access to services for people with hearing impairment?

•What is the access to hearing care for ethnic minorities?

Variation and guidelines:

What is the regional variability in practice in hearing aid provision for people with tinnitus and hearing loss? Assessing and addressing variation in tinnitus management.

Assessing the impact of guidelines for unilateral tinnitus on prognosis, outcomes and patient safety.

Should guidelines be developed for: the monitoring and management of ototoxicity; the management of sudden hearing loss?

Expanding current guidelines: should we reconsider thresholds for CIs; BAHAs; middle ear implants?

Commissioning and rationing:

Are we using the best selection criteria/assessments to determine candidature for hearing aids and what effect does this have on commissioning and meeting clinical need?

Should the criteria for funding cochlear implants and BAHAs be expanded?

What are the different levels of hearing assistance commissioned and available to cope with single sided hearing loss in different areas? Should routine aiding of mon-aural hearing losses be commissioned?

What is the extent to which the NHS is providing microsuction in different parts of the country?

Is there a benefit to a system of private or NHS contribution to commissioning hearing aids outside of the NHS?

Improving communication between healthcare professionals and patients:

What is the impact of professional expectation of outcomes of cochlear implantation, and how they are communicated to patients, on actual patient outcomes?

What is the impact on quality of life and social interaction of raising public awareness of the "hidden" conditions of hearing impairment and tinnitus and their impact on psychosocial wellbeing and interactions? Are patients receiving enough education regarding hearing and balance?

How can clinicians communicate uncertainty around the cause of tinnitus, its treatment and outcomes, and what impact does this have on the patient?

Communication

Improving communication between healthcare professionals: What strategies improve communication between healthcare professionals and what is their impact on patient safety and quality of care for tinnitus and hearing loss?

Communication between patients:

What is the impact of individual and group support networks on condition---specific and psychosocial patient outcomes for tinnitus, hearing loss and acoustic neuroma?

Paediatric and adult cochlear implant: how does the experience of adapting to a new method of analysing sound differ in different age groups and are there coping strategies that can be shared between groups?

Dissemination of research to the public:

What is the psychological impact of making current and planned research on hearing aids and tinnitus available to patients?

Decision-making and informed consent:

What is the most effective way of providing information on treatment options: type of BAHA and alternatives; treatments for acoustic neuroma? What is the benefit of shared decision-making tools for acoustic neuroma? Should we standardize consent for the treatment of acoustic neuroma?

Education

What is the impact on patient care of deaf awareness and communication tools training for healthcare professionals? What is the impact on audiological training of being academically grouped with purely diagnostic disciplines?

What is the impact of training GPs in testing to differentiate CHL and SNHL on patient care?

What is the impact of extending training for ENT surgeons and audiologists on patient management, outcomes and referral rates for single sided deafness?

Establishing a Research Culture

Research and technology: How can we utilise existing technology, such as hearing aids and mobile phones, to engage patients in research and collect real world patient data that will help us understand disease?

The global perspective: Can we develop effective medical treatments for sensorineural hearing impairment that can restore hearing and eliminate the need for hearing aids for the vast majority and thus provide a viable source of help for those in less---developed countries?

The Ageing Population

Understanding Health and Illness

Incidence and prevalence: What is the prevalence of hearing loss in the elderly in the UK?

Risk factors and triggers: What are the factors that increase the risk of developing age---related hearing loss?

Links to other conditions: Assessing ENT manifestations of Parkinson's and other neuro---degenerative disorders associated with age.

Pathogenesis: What are the mechanisms of hearing loss in middle age and in older age?

Indirect healthcare costs:

What are the socioeconomic impacts of balance disorders, including the impact of prolonged disability and falls in the elderly due to dizziness?

What is the societal impact of hearing and communication difficulties, including damage to voice, in older adults? What is the burden on family and carers of dizziness and falls in the older adult?

Assessing impact of ENT conditions in the older patient:

Determining the attitudes around ENT problems in the older adult: is this more than just "normal" aging?

Age-related hearing loss: what is its impact on social isolation, normal activities of daily living and independence? How do problems with balance contribute to poor quality of life in the older patient?

Impact on cognition and dementia:

Improve understanding of the link between hearing and cognition in the ageing adult.

Does intervention by means of hearing aid / cochlear implant result in slower progression of dementia, or reduced chance of developing dementia in older adults? (Look prospectively at the UK population of CI users over 60, and consider the impact of implantation on incidence of dementia).

Impact on work and employment: Is the impact on seeking employment greater in older patients with hearing loss than in younger patients?

Prevention of Ill Health, Maintenance of Good Health

Primary prevention: Developing primary prevention strategies for age-related hearing loss.

Secondary prevention:

Progressive hearing loss with age:

•What are the practical and cost-effective methods of developing an adult hearing screening programme for older adults/adults diagnosed with dementia?

•What other strategies can be developed to slow or delay the development of progressive hearing loss with age?

Age-related balance problems:

•What multidisciplinary screening models can be used to develop a screening programme for balance problems in the older adult?

•What is the impact of a programme on sequelae of falls and on the wider society?

Tertiary prevention:

Hearing rehabilitation:

•What is the impact of hearing rehabilitation for the older adult on cognition/social isolation/mental and physical health/quality of life?

•Would teaching sign to the over 50's aid communication? Vestibular rehabilitation:

•What is the role of vestibular rehabilitation for the older adult and where should it be delivered (primary care/community)?

•What is the impact of vestibular rehabilitation for the older adult on reduction in falls/costs to the NHS/acute hospital admissions/quality of life/maintaining independence/preventing social isolation?

Self-help strategies to maintain function:

•How do we encourage older adults to not accept loss of function as a normal part of aging and what impact does this have on health behaviours?

•How accessible is information on self-management that is available online/through mobile phones to the older adult?

Clinical Effectiveness

Diagnostic criteria: Research to develop investigation protocols to consider all causes of hearing loss/balance issues in the older adult so that diagnoses such as acoustic neuroma are not missed.

Timeliness and accuracy of diagnosis: Does early diagnosis and management of hearing loss in an aging population reduce risk of dementia?

Challenges in diagnosis:

How do perceptions that imbalance is a normal part of aging age affect diagnoses of vestibular dysfunction in older adults?

How can a GP/geriatrician/ENT consultant identify older adults with multiple comorbidities who may benefit from vestibular rehabilitation?

Timings of interventions: Does early hearing aid provision with mild or moderate age---related hearing loss enhance their value later on?

Adopting technology: What is the cost effectiveness of multi---channel programmable hearing aids for presbyacusis?

Developing novel therapies:

Developing medical therapies and appropriate delivery mechanisms for age related hearing loss.

Therapies for improving age-related loss of sense of smell and taste.

Research to eliminate ear infections in older adults.

Adapting existing interventions: How do we exploit the importance of cognition for hearing in future interventions, especially for older people and children? Can we use wireless devices, spatial hearing and brain monitoring as intervention strategies and trophic-support for dementia, hearing loss, tinnitus and 'auditory processing'?

Targeted therapy: Restoration of age-related hearing loss using gene therapy.

Designing and Delivering Healthcare

Patient pathways:

Balance pathways:

•What is the most cost---effective way to manage balance disorders in the older adult: how and where should they be delivered? Is there a role for specialised "dizzy clinics?"

•Which members of the multidisciplinary team should be involved in the assessment and treatment of older adults with balance problems?

•What is the impact of this pathway on falls, safety and continued independence?

Innovation and service delivery: How acceptable is the use of new technology (virtual clinics/online support/mobile phones) in organizing and delivering care to older adults?

Barriers to using services:

How can we improve access to hearing services for older adults who are housebound or in residential care, and for those with dementia?

What is the impact on hearing aid design on uptake: the need for dexterity with small devices?

Resources and an ageing population: How do we address growing demand for age-related ENT referrals?

Communication

Raising public awareness: What is the impact of raising public awareness of hearing impairment as more than a condition associated with age and infirmity and the impact this has on health behaviours?

Education

Medical Knowledge:

Is there recognition and knowledge of management of age---related balance problems in primary care?

What is the impact on patient care and outcomes of teaching healthcare professionals about the effect of age---related balance problems on quality of life, patient safety and health?

Balance Problems
Understanding Health and Illness

Understanding Health and Illness

Incidence and prevalence: What is the burden of disease for vertigo and Mal de Debarquement Syndrome (MdDS)?

Risk factors:

Are the following risk factors in causing or worsening the disorders: Meniere's disease and grains; viruses/infection or stress for balance disorders?

Causation:

What are the underlying causes for: Meniere's disease; balance disorders (general/vertigo; vestibular neuronitis/labrynthitis); MdDS?

Triggers: What are the triggers for vertigo and Meniere's disease (weather; hormones; stress; time of day)?

Links to other conditions:

Is there a link between acute vertigo and thromboembolic disease/stroke?

Is there a link between vertigo/Meniere's disease and neck pathology?

Are migraine, trigeminal neuralgia, Meniere's disease and chronic fatigue in some way linked? Does fibromyalgia have an effect on balance?

Pathogenesis:

What are the cortical processes/cognitive associates that lead to the perception of dizziness/vestibular disorders? What is the underlying disease process of Meniere's disease?

Genetics and heredity: Is Meniere's disease hereditary? Technology: Can animal models be used to understand vertigo? How conditions progress:

What are the natural history and outcomes of vertigo in the community?

Why do some people recover from unilateral vestibular insult better than others?

Indirect healthcare costs: What is the socioeconomic impact of balance disorders, including the impact of prolonged disability?

Impact of "hidden" condition: Understanding how having a "hidden" condition such as a balance disorder increases frustration and suffering due to the impact on social interaction, family interaction, interaction with healthcare professionals, enjoyment of life and external perceptions of intelligence.

Impact on functioning:

Assessing the disruption of balance problems to a "normal," healthy life and being able to engage in "normal" activities; understanding the degree of fatigue caused by balance dysfunction. What are the levels of distress/daily suffering caused by vertigo/MdDS?

Qualitative studies on patient experience of vertigo as in using our quantitative questionnaires, we may be missing some important dimensions of patient reported vertigo symptoms and sequelae.

Psychosocial impact: Assessing the impact of Meniere's disease on social isolation and withdrawal.

Fear and safety:

Understanding the patient experience of fear that vertigo will recur and uncertainty over managing acute episodes. Understanding the patient experience of fear of falling due to imbalance.

Impact of delay: What is the impact of long waiting lists on quality of life and mental health for patients with long-term vestibular and balance problems?

Impact on work: What is the Impact of balance problems or Meniere's disease on ability to work?

Prevention of Ill Health, Maintenance of Good Health

Primary prevention: Are there any prevention strategies that can be used to avoid balance problems?

Secondary prevention: What effective secondary prevention strategies can be used to prevent the progression of Meniere's disease?

Tertiary prevention:

Vestibular rehabilitation:

•What is the role of self-care in vestibular rehabilitation for chronic vestibular disorders?

•Develop evidence of effectiveness/effective strategies by assessing the impact of rehabilitation on quality of life/mental health/social interaction/maintaining independence.

Is there a role for tertiary prevention of Meniere's disease in reducing or preventing relapses, improving quality of life and promoting strategies for independence?

Clinical Effectiveness

Diagnostic testing:

Do functional tests (i.e. balance function testing) better reflect real life patient difficulties as opposed to objective tests?

Vestibular testing:

•Develop more sensitive tests for diagnosis of vertigo/balance disorders

•Consider less invasive alternatives to caloric testing such as vHIT, VEMPs.

•What is the efficacy of the tests that we use in diagnosing balance problems (including VNG, caloric, posturography, and VEMPS)?

•Assessing which specific vestibular membrane frequencies are affected to target vestibular rehabilitation.

•Assessing cognitive processing and spatial cognition as part of dizziness and imbalance assessment.

•Accurate diagnostics for vestibular migraine.

Diagnostic criteria:

Meniere's Disease:

•Develop agreement on the primary and secondary characteristics for diagnostic purposes and agreement on the basic definition of terms.

•Distinguish the underlying illnesses in what are a wide range of different patient experiences currently gathered together under the portmanteau description "Meniere's disease and Meniere's variants."

•Timeliness and accuracy of diagnosis:

•What is the effectiveness of early, correct diagnosis on outcome (quality of life/mental health) in patients with balance/vestibular dysfunction?

•Design a survey to diagnose the nature of a balance problem on---line to avoid delay in diagnosis caused by oversubscribed ENT clinics.

•Timing of interventions: What is the effect of early, correct diagnosis and clinical treatments/interventions, including neurophysiotherapy and access to information about other patients' experience, for vestibular function/ balance rehabilitation and whether these speed up the process of recovery?

•Assessing interventions:

•Does the Epley manoeuvre for benign positional vertigo work?

•What is the evidence for a wider application of vestibular rehabilitation in large numbers of adults with balance problems?

•Are exercises for balance and vertigo effective?
•Are oral pharmaceutical treatments for vertigo effective (prochlorperazine/cinnarizine)?
•What is the evidence for current treatments for vestibular migraine?
•Do intratympanic steroids actually work in Meniere's?
•Is salt restriction really necessary in Meniere's disease?
•What is the best treatment for dizziness/vertigo/chronic vestibular imbalance; which vestibular rehabilitation exercises are most beneficial?
•Which migraine treatments work best for vestibular migraine?
•Improving existing interventions:
•How do we optimise symptomatic control and rehabilitation to restore quality of life and independence for patients with dizziness and imbalance?
•Better treatments for atypical migraine causing dizziness and facial pain.
•Selecting patients:
•What are the indications for referral for vestibular rehabilitation for patients with Meniere's disease?
•Who would benefit from CBT combined with standard vestibular rehabilitation?
•Is there any benefit from vestibular rehabilitation to patients with longstanding balance problem due to acoustic neuroma removal?
New therapies:
Create treatments for Mal de Debarquement Syndrome/Meniere's disease/balance disorders or find ways to put
patients into remission. Development of a vestibular implant.
Is there a role for stem cell therapy in restoring lost balance function?
Identify subgroups to customise treatment:
Understand better the 'incomplete compensation' group of vertigo patients to develop customised treatment rehabilitation programmes.
Distinguish the underlying illness described as Meniere's disease and variants to determine treatment.
Outcome measures:
Develop condition specific quality of life outcome measures for dizziness.
Designing and Delivering Healthcare
Patient pathways:
"Dizzy clinics":
•What is the need for and effectiveness of balance clinics or specialised balance centres? What is the impact of specialised clinics on morbidity and quality of life?
•What MDT would be needed for these clinics? Would an audiology-led clinic be safe for patients?
•Where should these clinics take place: community or primary care-based clinics?
•What would be the access and referral pathway to such clinics?
What are effective strategies or pathways for acute vestibular symptoms/attacks to avoid presentation to A&E? How effective are online or mobile phone programmes for vestibular exercises?
How do you assess and incorporate diversity in presentation and response to treatments and interventions (Meniere's disease; vertigo) to patient pathways?
What is the impact of audiology-led triage for balance and dizziness issues on ENT capacity problems and waiting times?

Access to services: Is it cost-effective to provide balance assessments in local clinics for patients who find it difficult to travel due to their balance problems?

Communication

What is the impact on patient outcomes of clinician belief in the efficacy of vestibular rehabilitation? How can this option be communicated to patients so that it has a positive impact on their outcomes?

What are the strategies that can be effectively employed to improve listening and communication skills? How do we acknowledge patients' symptoms and experience of disease, and address patient concerns about Meniere's disease?

What is the impact on patient safety of raising public awareness of balance conditions?

Education

What is the impact on patient care, safety and referral rate of training GPs in diagnostic tests and treatments for vertigo (Dix---Hallpike; Epley; the role of vestibular sedatives)?

What is the impact on developing communication and interpersonal skills of healthcare professionals by raising awareness of the effect of Meniere's disease on the patient?

What is the impact on patient care and outcomes of teaching healthcare professionals about the effects of Meniere's disease on quality of life, patient safety and health?

Is there recognition and knowledge of Meniere's disease and vertigo in primary care?

What is the impact of extending training in vertigo for ENT surgeons and audiologists on patient management, outcomes and referral rates?

Establishing a Research Culture

What is the value of research into common conditions that have not traditionally been prioritised such as balance? Collaborate with neuroscientists and neurologists to research vestibular migraine and dizziness.

Children and Young People

Understanding Health and Illness

Incidence and prevalence:

What is the incidence and prevalence of OME (glue ear) in general paediatric population and in certain demographic populations (ethnicity)? Why is the prevalence of OME higher within certain demographic populations (ethnicity) in the UK?

•What is the prevalence of unmanaged hearing loss in older children immigrating to the UK and how should this be managed?

•What is the prevalence of APD and how does it compare to prevalence of deafness in children?

•What is the prevalence of tinnitus in different age groups?

•What is the prevalence of paediatric vestibular/balance disorders?

•What is the incidence of hearing impairment in children compared to diagnosis rate?

Risk factors and triggers:

•Determining the effect of diet (especially dairy) in developing nasal congestion, glue ear and suppurative otitis media in children.

•Determining the role of viruses and infection in childhood hearing loss.

•What is the risk of future hearing loss from young people using personal listening devices?

•Is the use of mobile phones a risk factor for future development of acoustic neuroma?

•What are the risk factors for an individual developing allergies?

Links to other conditions: what is the link between allergic upper airways disease and lower airways disease, and how do you reduce the risk of progressing to the latter?

Pathogenesis: understanding neurological variants of laryngomalacia.

How conditions progress:

•What is the prognosis of children with congenital CMV who at birth are only symptomatic with hearing loss?

•Understanding the natural history of retraction of the tympanic membrane and development of cholesteatoma; natural history of cholesteatoma in adults and children.

•What is the prognosis of ANSD?

•Is the presence of middle ear congestion at time of newborn hearing screening tests an indicator of longer-term issues of glue ear?

•Long-term outcome study for children with glue ear, including impact on middle ear.

•The effect of recurrent tonsillitis on the upper respiratory tract; long-term impact and complications of recurrent ENT infections, including middle ear infections.

Indirect healthcare costs: what is the societal impact of hearing and communication difficulties (including damage to voice) in children?

Impact on daily functioning: What is the impact of anosmia on eating habits and wellbeing of adults and children?

Developing coping strategies: Assess patient experience (both paediatric and adult) following cochlear implant. Can the different experiences of different age groups in adapting to a new method of analysing sound be used to develop coping strategies that can be shared between groups?

Impact on learning and development:

•Long-term impact of glue ear or other types of childhood hearing loss on learning and cognitive function of children.

•Does auditory processing disorder have an impact on academic achievement? Can this be improved by quieter classrooms?

•What effect does mild hearing loss have on auditory memory and does this have an impact on educational attainment?

•What is the impact of fluctuating hearing loss on speech and language development? Does delay in treatment/lack of treatment of glue ear in young children lead to long---term damage to speech? Can the difficulties in being understood in early childhood have severe consequences to educational, language and emotional development?

•The impact of allergic rhinitis, recurrent tonsillitis and OSA on educational attainment in children.

•What is the impact of vestibular hypofunction on children's learning, particularly if they have lost function or if they have additional needs? If a significant vestibular hypofunction is identified early can we help to minimise the effect on the child's learning and locomotor ability?

•The effects of tongue tie on speech, language and feeding.

Prevention of Ill Health, Maintenance of Good Health

Develop and implement primary prevention strategies for:

•Glue ear in: normal population; children with Down's syndrome or cleft palate; children from ethnic demographic populations with higher prevalence.

•What effect has the pneumococcal vaccination programme had on childhood hearing loss?

•Subglottic stenosis secondary to intubation.

Secondary prevention:

•Is there benefit of immunotherapy early in the treatment of allergic rhinitis to prevent progression of the allergic march in children?

•How do we prevent recurrent ear infections in children?

Tertiary prevention: ANSD (Auditory Neuropathy Spectrum Disorder) rehabilitation in children: What interventions (Audiovestibular therapy; support at school) maximise potential? What is the impact of early language intervention on reducing language difficulties and learning difficulties during school years?

Clinical Effectiveness

Diagnostic testing and assessment of function:

Development of an Auditory Steady State Response protocol for diagnostic hearing assessment of a baby hearing post--- screening

•The reliability of cortical evoked potentials to indicate hearing in infants at screening levels

•Determine age---related correction factors and data on interaural attenuation to improve accuracy of ABR testing.

•What methods can be used to assess hearing thresholds in young babies diagnosed with ANSD before they are old enough to carry out reliable VRA testing?

•Vestibular/balance testing in children: is it needed for all children with motor co---ordination difficulties or only those with hearing problems?

•Role of diagnostic aspiration in AOM.

Diagnostic criteria:

•What is the optimal set of guidelines for assessing children with tinnitus?

•What is the optimal set of guidelines for assessing children with balance problems?

•Diagnostic tool for APD; how to distinguish children with APD from those with learning problems.

•Is there benefit to using the ECLiPS questionnaire to assess children with glue ear prior to using hearing aids?

•Diagnostic criteria to differentiate tonsillitis from sore throats.

•Standardise evaluation and diagnostic criteria for OSA.

Timeliness and accuracy of diagnosis:

•Is there any impact on outcome for children with vestibular impairment by identifying them with vestibular testing?

•How often does hearing impairment in children go undiagnosed?

•How to diagnose neurological variants of laryngomalacia sooner in order to improve detection and subsequent treatment of other neurological symptoms.

Challenges in diagnosis:

•The role of multidisciplinary assessment and feedback, including parents and carers, in the diagnosis and management of hearing loss in children, especially in the context of children with additional support/ complex needs.

•Under diagnosis of ANSD due to lack of awareness of the condition.

Safety, complications and deterioration:

Management of recurrent tonsillitis:

•Does reducing the number of tonsillectomies performed have an impact on health in adulthood?

•Assess the impact of reduction in number of tonsillectomies on hospital admissions for tonsillitis and its complications.

•Which antibiotic should be used for acute episodes and how long should the course last?

Glue ear: Does the fluid in glue ear cause permanent damage to the middle ear and if so do grommets prevent the damage? ENT infections:

•What more could be done to prevent serious complications of ENT infections (mastoiditis; acute sinusitis and the early use of decongestants; meningitis; eardrum perforations; the use of simple intervention such as hand hygiene)?

•Can prophylactic antibiotics help prevent frequent ear infections? Does inaction have long term effects that are more significant than in adults to the child's developing anatomy, physiology and neural plasticity?

Timings of interventions:

•Is there any benefit from introducing early physiotherapy in the first year of life for those with congenital vestibular areflexia? What is the benefit of physio in the early years for this cohort or for those who lose vestibular function? If a significant vestibular hypofunction is identified early can we help to minimise the effect on the child's learning and locomotor ability?

•Determining the effects of tongue-tie on speech, language and feeding development in children, and timing/effectiveness of intervention.

•What is the impact on outcome of developing communication strategies for children with significant hearing impairment?

•Success rate of tolerating and effective use of hearing aids in the under 5's.

•What is the best method and timing to manage consistent hearing loss and fluctuating hearing loss in children under 5 years?

•Does the early and continuing placement of grommets aid the development of normal speech patterns, reducing long-term damage to speech and impact on educational, language and emotional development?

Adopting technology: How best to improve the classroom acoustics or provide FM systems for children with APD.

Improving interventions:

Improve assessment of benefit of hearing aids and cochlear implants in infants:

•Can CAEPs reliably indicate access to sounds using hearing aids (reliability of CAEP measures; the effect of hearing aid processing on the CAEP test stimuli)?

•Can electrophysiological measurements reliably indicate uncomfortable loudness levels when fitting hearing aids in infants? Can parameters from EPs reliably indicate loudness growth and hence be used to set compression functions in hearing aids (in particular for infants)?

Consider combined interventions in the elimination of biofilms.

Selecting patients for interventions:

Children with glue ear: do grommets confer benefit on long---term educational outcomes/speech/hearing? If so what are the selection criteria/threshold/age to intervene?

Would children with severe hearing loss do better with regular speech and language therapy input? Hearing aids:

•Develop the evidence to determine who would benefit from a hearing aid.

•What are the benefits of providing hearing aids for mild---moderate hearing loss? Cochlear implants:

•Is there a role for cochlear implants in children with moderate hearing loss not at present eligible?

•Would children with severe hearing loss do better with cochlear implants than hearing aids?

•What is the indication for cochlear implants in ANSD?

What is the benefit of tonsillectomy/adenoidectomy in disabled children with upper airway difficulties?

Assessment of current interventions:

Paediatric hearing disorders:

•Assessing the effectiveness of current interventions for ANSD and APD (provide better information for parents).

•Glue ear: the long-term impact of current treatments and impact on learning/cognitive functioning/educational outcomes/speech/hearing (grommets). Assessing the role of non-conventional treatments in the management of glue ear.

•Success rate of tolerating and effective use of hearing aids in the under 5's.

Which Eustachian tube treatments are effective in the long term for treatment of glue ear and CSOM in children and adults? What is the effectiveness of surgical intervention in children with tongue-tie?

Assessing the efficacy of paediatric tonsillectomy:

•For recurrent tonsillitis, including impact on educational attainment.

•Outcomes of adenotonsillectomy for obstructive sleep apnoea in children.

•Cost-effectiveness of tonsillectomy.

•Assessing the effectiveness of current management of laryngomalacia.

•How effective is the current management of allergic rhinitis in children?

•Develop evidence for tracheostomy decannulation protocols; impact of decannulation protocols on time to discharge (adult and paediatric).

Assessing antibiotic use:

•In upper respiratory tract infections/conditions.

•Role, type, duration of antibiotics for AOM.

•Recurrent AOM and antibiotic sparing approaches: is there a role for grommets?

Comparing interventions:

Grommet vs hearing aids:

•The benefits of hearing aids vs grommets for children with glue ear (long term benefits to hearing, speech and language development, development; impact on child's developing anatomy, physiology and neural plasticity).

•Are grommets or hearing aids more beneficial for patients with cleft palate and hearing problems?

•What is the best treatment approach to the management of glue ear in children with Down syndrome: hearing aids or grommets (or t-tubes)?

What are the best treatments for glue ear and suppurative otitis media that can be delivered in primary care? What is the best vestibular rehabilitation approach for children with vestibular hypofunction?

Developing novel therapies:

Conditions where new, effective therapies are needed: APD; ANSD; balance disorders (paediatric BPPV and migraine treatment) Developing new biological treatment for congenital hearing loss. Develop treatments for glue ear.

Develop therapies for congenital lymphangiomas; the use of Viagra. Research to eliminate ear infections in children/older people.

Adapting existing interventions:

Should children with APD be encouraged to learn musical instrument?

How do we exploit the importance of cognition for hearing in future interventions for children: includes using wireless devices, spatial hearing and brain monitoring as intervention strategies and trophic---support for dementia, hearing loss, tinnitus and 'auditory processing'.

Would a slow release drug eluting implant help treat the children with CSOM?

Targeted therapy:

Study with adequate numbers and ENT/hearing assessment to establish when cows milk protein intolerance causes or worsens serous otitis media and nasal/sinus congestion: if there is a link are there any biological markers (possible role of component assays)?

Developing outcome measures:

Use consensus-based approaches to develop outcome measures, especially to determine what domains of impairment are important, for OME (glue ear) and 'routine' surgical interventions in children.

Develop multidisciplinary assessment tools with easily recordable and rapidly shareable functional outcomes to inform ongoing hearing rehabilitation in children.

Is there evidence to supports speech audiometry as a predictor of hearing aid outcome? Develop more functional outcomes measures for hearings aids than PTAs for adults and children.

Designing and Delivering Healthcare

Patient pathways:

•How do we develop age-appropriate pathways in hearing impairment to account for the impact on development and opportunities for children and young people?

•Considering patient/parent choice in the implementing of clinical services in regard to management of glue ear, and how this is affected by the interactions and input of the MDT.

•How do we design a pathway to allow for the diagnosis and management of hearing loss in children with complex needs/requiring additional support?

•What is the benefit of developing paediatric tinnitus clinics combining tinnitus and hyperacusis management?

•Developing integrated care pathways for glue ear: Which outcomes are most important (agreement) and how can these be used in developing a rational/ good chronic disease model of care in OME; Can we provide better community---based multi---disciplinary team for diagnosis and care of OME in children; Developing a better model of care to reduce strain on paediatric audiologists, educational audiologists and teachers of the Deaf.

Developing paediatric balance pathways: The impact of designing and developing paediatric balance pathways on outcomes; Assessing current services and unmet need; What services are available for vestibular assessment and how do they look/compare?; Should we combine paediatric balance and hearing pathways?

•How do we develop a tracheostomy pathway for children: time to decannulation and time to discharge? Where to deliver service: How could audiology---led services be made more accessible to children and young people? Access to services and technology:

•Improving or standardising the recognition of childhood hearing loss in community services.

•Is it cost effective for schools to fund personal FM systems or sample systems for children with APD?

The sustainability of services: The impact of clinical advances in diagnosis/management in child hearing loss and increasing parental expectation on the sustainability of multi-specialist services despite diminishing resources.

Guidelines:

•Developing guidelines in assessing paediatric tinnitus

•Developing guidelines for managing APD

•Guidelines for antibiotic use in upper respiratory tract infections

•Assessing the evidence base and quality of current NICE OME (glue ear) guidelines especially with regard to long-term outcomes.

Commissioning:

•Is using the PTAs the best assessment to determine which children will benefit from grommets for glue ear, and what effect does this have on commissioning and meeting clinical need?

• Should a multidisciplinary assessment for children with APD be commissioned in view of comorbid conditions?

Communication

Communication between patients: Paediatric and adult cochlear implants: how the experience of adapting to a new method of analysing sound differs in different age groups and if there are coping strategies that can be shared between groups.

Helping patients make decisions: Assessing and improving existing shared decision---making instruments for management of glue ear.

Education

The impact of extending training for ENT surgeons and audiologists on patient management, outcomes and referral rates for velopharyngeal reflux and paediatric balance disorders.

Are paediatricians being given sufficient training in ENT in their postgraduate training?

How can paediatricians improve their knowledge about ear problems in order to improve patient care?

Establishing a Research Culture

Developing a culture of value in research:

How do we determine the comparative values of research and interventions in paediatric ENT procedures compared to other ENT and non---ENT procedures?

Head, Neck and Airway

Understanding Health and Illness

Impact on daily functioning: What is the impact on patients of poor functional outcomes for speech and swallowing following head and neck cancer treatment?

Social impact: What is the impact of facial disfigurement on social isolation and withdrawal?

Incidence and prevalence:

What is the burden of disease of globus pharyngeus?

What is the incidence of subglottic stenosis secondary to intubation?

Risk factors: What risk factors in the individual that predispose them to choking and problems with swallow?

Prognosis: What is the life expectancy and mortality from respiratory pathology in people with learning disabilities who have eating, drinking and swallowing problems?

Causation: What are the underlying causes of globus pharyngeus/snoring/obstructive sleep apnoea? Pathogenesis: What is the disease process of head and neck cancers; how does HPV lead to oropharyngeal cancer? Genetics and heredity: Is cancer of the tongue hereditary?

Links to other conditions:

Can hearing be affected by a laryngectomy?

Does surgery for OSA reduce cardiovascular morbidity and mortality? What is the impact of OSA on general health? What are the links between bruxism, laryngopharyngeal reflux, stress and globus pharyngeus?

Is there a link between hearing loss and snoring?

Relationships: What is the effect of snoring on intimate relationships?

Safety: What is the risk to safety of patients with OSA due to road traffic accidents?

Work:

What is the impact of OSA on current employment?

What is the impact of dysphonia on future opportunities or seeking employment?

Prevention of Ill Health, Maintenance of Good Health

Primary prevention:

Vaccination and head and neck cancer; what is the role of universal HPV vaccination? How can subglottic stenosis secondary to intubation be prevented?

Secondary prevention:

Head and neck cancer screening programmes:

•Is it feasible to develop a screening programme?

•What impact could new optical imaging techniques have on a screening programme?

•Tertiary prevention: Self-management of snoring: can people use strategies for quieter sleeping?

Clinical Effectiveness

Diagnostic testing:

What is the role for improve diagnostics for endoscopy (eg confocal imaging, contact endoscopy, narrow band) in head and neck cancer?

How can emerging diagnostic technology be used in screening for head and neck cancer?

Diagnostic criteria:

What are the criteria for evaluation of neck lymph nodes in primary care? What are the diagnostic criteria for laryngopharyngeal reflux?

How do we standardise the evaluation and diagnostic criteria for OSA?

Timeliness and accuracy of diagnosis: How do we detect or diagnose head/neck tumours earlier?

Existing therapies:

How can speech rehabilitation post laryngectomy be improved, especially for women?

How can we improve the effectiveness of electrical stimulation of the upper airway dilator muscles in obstructive sleep apnoea to provide alternatives to CPAP?

How can we improve surgery results for OSA?

Is there a link in reduction in the number of tonsillectomies and an increase in oropharyngeal cancer of tonsillar origin?

Safety and complications:

•How can we minimise post chemoradiotherapy complications in head and neck patients?

•How safe are laser resection margins in laryngeal cancer?

•What is the evidence base for drains following surgery and for how long we should leave them in? What is the best method of induction of anaesthesia in the obstructed airway?

What improvements can be made in tracheostomy tube design to reduce or eliminate accidental decannulation or blockage?

Does surgery for OSA reduce cardiovascular morbidity and mortality? What is the effectiveness of current surgical management of OSA?

Develop evidence for tracheostomy decannulation protocols; impact of decannulation protocols on time to discharge (adult and paediatric).

What is the evidence and effectiveness of Vocal Hygiene Advice to patients with voice disorders (evidence for post surgical voice rest, dietary advice, use of PPIs)?

Management (Investigation and treatment) protocol for globus pharyngeus. How effective is current management of cricopharyngeal spasm/symptoms? What is the benefit of lansoprazole compared with placebo for globus pharyngeus?

Timings of interventions: Could earlier detection of glomus jugulare tumours mean non---surgical options, or less invasive surgical options with fewer complications, are possible?

Adopting technology: Should ENT units be investing in robotic surgery? What will be gained for patients?

Improvement of existing interventions: How do we improve outcomes from head and neck cancer?

Assessing current interventions:

•The efficacy of neck dissection in recurrent squamous cell carcinoma of the head and neck.

•What is the optimum timing of intervention and type of SLT intervention for swallowing, speech and voice problems in people who have had radiotherapy and/or chemotherapy?

•The effectiveness of current SLT practice for patients with laryngectomies.

•Utility of level IIb dissection in regionally-metastatic papillary thyroid carcinoma.

•Management of Micropapillary (< 1 cm) thyroid carcinoma.

•The role of radioiodine treatment in minimally invasive thyroid carcinoma (follicular/follicular variant of papillary): do these need further treatment other than lobectomy?

•Management of dysplasia.

•Can HPV positive head and neck SCC treatment be de-escalated?

•Head and neck cancer: what is the quality of life for surgical vs non-surgical treatments?

Stapling of pharyngeal pouches: what is the long-term outcome? Have we wrongly moved away from open surgery, particularly in younger patients?

What is the comparison of concurrent chemoradiotherapy with surgery plus postoperative radiotherapy on locoregional failure and disease-specific survival in locoregionally-advanced orophayrngeal and laryngeal squamous cell carcinoma?

New therapies:

Develop drugs with minimal side effects to reduce throat/stoma mucus for laryngectomy patients.

Voice reinstatement in laryngectomees: utilising advances in computing technology and materials science; research into the development of a 'Voice amplifier' small electronic device that could be implanted into the throat.

Consider the development of an artificial windpipe to the mouth area, which would help alleviate some of the mucus problems that Laryngectomees face today, and do away with neck breathing.

Targeted therapy: Develop techniques of genetically-informed diagnoses for ENT conditions and head and neck cancers to personalise the therapeutic approach; determine the cost-effectiveness of these diagnoses.

Work with molecular scientists to develop targeted molecular therapies for benign ENT conditions and head and neck cancers.

Outcome measures:

Develop condition-specific quality of life outcome measures for head and neck cancers.

Develop functional outcome measures for throat cancer, including outcomes for voice and swallow. What condition specific quality of life outcome measures should be used for globus pharyngeus?

Designing and Delivering Healthcare

Patient pathways:

Can we standardise a pathway for the management and referral of neck lymph nodes from primary care? Can we standardise the pathway for prolonged dysphonia?

What is the impact of delay in referral on patient health, well-being and quality of life in dysphonia? How can the pathway for prolonged dysphonia be standardised?

How do we establish a defined treatment pathway for snoring?

Tracheostomy pathways: determining numbers of patients, practice, tube changes, strategies for decannulation, protocols for accidental decannulation and MDT members involved in the pathway.

Pathway for functional dysphonia:

•Is there unmet need for management of functional dysphonia?

•How do we develop a model for this pathway?

Access to services: What is the need for SLT assessment and treatment in people with learning disabilities, and how do we improve their access to this service?

Safe follow up: What is the optimal strategy for follow up after head and neck cancer treatment including the potential for costs savings by nurse-led follow up and frequency of follow up?

Practice variation and guidelines:

Assessing and addressing variation in the management of micropapillary thyroid carcinoma and laryngeal dysplasia. Assessing and addressing variation in the management of globus pharyngeus.

What would be the impact of developing guidelines for laryngeal dysplasia?

What is the impact of guidelines for unilateral nasal polyps on prognosis, outcomes and patient safety? How do we develop guidelines for globus pharyngeus?

Commissioning: Assessment of different commissioning and availability for robotic head and neck surgery.

Communication

Assessing communication in MDT settings and its impact on quality of care.

Consenting patients for head and neck cancer treatments and procedures: how well do we communicate information on treatment, side effects and outcomes in head and neck cancer to our patients to enable them to make an informed decision?

Assessing the impact of published surgeon-level outcome data on consent and decision-making.

Education

What is the impact on patient care and outcomes of teaching healthcare professionals about the effect of laryngeal dystonia on quality of life, patient safety and health?

Mental Health and Dementia

Understanding Health and Illness

Pathogenesis: Understanding the mechanisms behind auditory processing: can improving cognition improve hearing?

Psychosocial impact:

Assessing incidence and severity of depression/"mental health problems" in patients with hearing loss, tinnitus, vestibular disorders, Meniere's disease, CRS, anosmia, loss of sense of taste, facial disfigurement, MdDS, hyperacusis. Are patients with tinnitus, Meniere's disease or MdDS at increased risk of suicide?

How do we determine when patients require psychological support in the above conditions?

Improve understanding of the link between hearing and cognition/ageing. What is the association between hearing loss and dementia?

Does early diagnoses and management of hearing loss in aging population reduce risk of dementia?

Does early management of hearing loss in patients with dementia delay their admission in to residential care? Would screening/fitting hearing aids at an earlier stage influence the pathway of dementia? Does intervention by means of hearing aid / cochlear implant result in slower progression of dementia, or reduced chance of developing dementia? (Look prospectively at the UK population of CI users over 60, and consider the impact of implantation on incidence of dementia).

What would be the impact of an adult hearing screening programme and hence earlier aiding have on the development and progression of dementia?

How does dementia affect assessment and treatment for hearing loss?

Does improving use of hearing aids the results of using them have an impact on dementia? Why are cognitive issues important in vestibular disorders?

What are the cognitive aspects of vestibular illness, particularly their impact on confusion and memory loss?

What are the mechanisms behind auditory processing and how this can affect a person's cognition? Can improving cognition improve hearing?

Coping strategies: Determining when and how to provide psychological support for anosmia, acoustic neuroma, tinnitus and hearing and balance problems.

Impact of delay: What is the impact of long waiting lists on quality of life and mental health for patients with long--- term vestibular and balance problems or anosmia?

Work: Understanding the frustration of patients who are unable to work due to their ENT issues.

Prevention of Ill Health, Maintenance of Good Health

Secondary prevention: What is the impact of developing an adult hearing screening programme for adults newly diagnosed with dementia?

Tertiary prevention:

What is the impact of vestibular rehabilitation on mental health? What is the impact of hearing rehabilitation on cognition?

Should psychological support be offered with rehabilitation?

Clinical Effectiveness

Timeliness and accuracy of diagnosis:

What is the effectiveness of early, correct diagnosis on outcome (quality of life/mental health) in patients with balance/vestibular dysfunction?

Does early diagnosis and management of hearing loss in aging population reduce risk of dementia?

Challenges in diagnosis: How does dementia affect assessment and treatment for hearing loss?

Existing therapies:

When is it best to start hearing amplification to prevent decline in cognition due to hearing loss? What is the impact of hearing aids on cognition?

Designing and Delivering Healthcare

Patient pathways:

Should tinnitus patients be assessed for depression and referred to mental health teams as part of their pathway? How do we develop a "joined-up approach" to the assessment, diagnosis and management of dementia for people with hearing loss and deaf people who use BSL?

How can the need for emotional and psychological support be incorporated into the pathway for chronic rhinosinusitis and anosmia?

Access: How do we improve access to hearing services for patients in residential care and for those with dementia?

Capacity for services: What is the impact of dementia on assessment and treatment of hearing loss and what additional resources does audiology require to provide this service?

Communication

What is the impact of increasing awareness in healthcare professionals of the emotional and psychological impact of conditions on their communication with patients?

What is the impact of raising public awareness of "hidden" conditions and their effect on psychosocial wellbeing and interactions (hearing impairment; anosmia; allergy; tinnitus; balance)?

What is the impact of individual and group support networks on psychosocial outcomes for patients?

What is the psychological impact of making current and planned research available to patients (anosmia; hearing aids; tinnitus)?

Patient-Centred Care

Understanding Health and Illness

Understanding disease to improve care:

How can we use technology to understand causation of disease?

What is the long---term impact of loss of special senses on human health?

Work: Understanding the frustration of patients who are unable to work due to their ENT issues.

Prevention of Ill Health, Maintenance of Good Health

Tertiary prevention: How do we determine what information is needed for self---help strategies for patients with chronic conditions, and how do we deliver it, making greater use of information technology?

How can rehabilitation programmes be used to provide coping strategies for individuals and families and promote social interaction and communication strategies?

Clinical Effectiveness

New technologies and therapies:

How can we work with molecular scientists to develop targeted molecular therapies for benign ENT conditions? How do we develop an automated audit database for ENT operations: how can we datamine this to establish normative clinical trajectories and risks to build our knowledge and make our decision-making smarter and more personalised?

Outcome measures:

How can we incorporate validated quality of life metrics as outcome measures in main ENT conditions?

What consensus-based approaches can we use to develop outcome measures, especially to determine what domains of impairment are important?

Designing and Delivering Healthcare

Patient pathways:

Timely diagnosis and referral: How can GPs be supported in making ENT diagnoses and appropriate referrals: providing online guidance and referral tools; developing advice networks and awareness of local referral pathways? Is there a role for desktop advisors or apps for communication and referrals?

Assessing and improving service:

•How can we develop outcome measures to assess quality of service based on patient safety?

•Would using remote technology for appointments and follow up improve service and patient care?

•How do we assess the quality on ongoing care in ENT, Hearing and Balance care?

•What is good/best practice for treatment and follow up for patients with multi-sensory deficits?

•Pathways for patients with complex health needs: Establish what is good/best practice for treatment and follow up for patients with multi-sensory deficits (hearing/balance/vision).

Treating patients in primary care and the community: How acceptable is the change of location of care to patients?

Practice variation and guidelines:

What is the patient perception of practice variation?

How do we implement and increase compliance with guidelines in primary and secondary care?

How can ENT surgeons best influence management of conditions treated predominantly by other specialties (especially general practice) and thus reduce practice variation?

What does high quality ENT care look like and how can we reduce inappropriate variation within the system? What is the benefit of standardisation of practice versus flexibility to patient care?

Which areas in ENT currently require the development of high quality, high level prospective multi-centre clinical research which will result in the development of gold standard guidelines capable of changing practice?

Commissioning:

What are the consequences of decommissioning procedures for patients, including impact on current and future health?

Communication

Improving communication between healthcare professionals and patients: What is the level of patient satisfaction with current communication strategies?

How do we increase awareness in healthcare professionals of the emotional and psychological impact of conditions to improve communication?

What is the impact of raising public awareness of ENT conditions on clinicians acknowledging patients' concerns? What is the impact of patients educating healthcare professionals on patient-centric communication strategies? What is the impact of raising public awareness of ENT conditions on perceived and actual stigma of patients?

How do we provide information to patients about care pathways and expected follow up; what is the role of IT in communicating about service?

Communication between healthcare professionals:

What strategies improve communication between healthcare professionals and what is their impact on patient safety and quality of care; how do we adopt models of good communication and use technology to improve communication between primary and secondary care?

Communication between patients:

How do we increase awareness of existing support networks for patients, their families/carers and healthcare professionals?

What is the role of online support groups versus face-to-face support groups: what is the difference in impact on patients' health outcomes?

What is the role of developing protected patient forums in the NHS?

Managing uncertainty:

What are effective methods of communicating uncertainty around cause of disease, treatment and outcomes, and the impact of this on patients?

Education

How do we educate clinicians on the holistic management of complex patients? What impact does this have on patient safety and outcome?

Are GPs and paediatricians being given sufficient training in ENT in their postgraduate training? How much ENT is taught at the undergraduate level and should this be expanded?

Are nurses being given sufficient training in ENT?

Establishing a Research Culture

Value in research:

Develop research in areas (including rare conditions) that give added value by providing insight and information into other conditions.

Recognising the value of, and considering research into rare conditions: dystonia and swallow/speech/breathing; Mal de Debarquement Syndrome; congenital lymphangioma; ENT manifestations of Hunter Syndrome.

Recognising the value of, and considering research into common conditions that have not traditionally been prioritised: tinnitus; hearing loss; rhinosinusitis; anosmia; globus pharyngeus; balance; swallowing problems in learning disability.

Collaboration and engagement:

How can industry aid in putting support and processes in place to facilitate research and applying for funding? How do we engage with clinicians who are conducting informal research and provide structure to support and improve the value of their research?

How do we engage surgeons and put processes in place to encourage them to run properly funded studies?

How do we train, engage and support trainees in collaborating in high quality research, and how do we embed this in their formal training? What is the role of trainee collaboratives? What is the impact of involving trainees on the success of studies?

What are the non-clinical academic disciplines with whom we should be collaborating: innovations experts; bio-engineers; neuroscientists; implementation scientists?

What are the clinical specialties with whom we should be collaborating and how do we engage with them: primary care collaboratives; physiotherapists; gastroenterologists; paediatricians; respiratory physicians; high profile specialties?

Inflammation, Infection and Allergy

Understanding Health and Illness

Incidence and prevalence:

How do we use electronic health records or CPRD to evaluate the disease background of chronic rhinosinusitis in primary care?

What is the burden of disease for:

•Anosmia +/- taste disturbance?

•Chronic rhinosinusitis?

Why is the incidence of hayfever increasing?

Risk factors:

Does eating dairy increase the risk of developing suppurative otitis media?

Is diet, in particular dairy products, a risk factor for developing rhinosinusitis or anosmia? Do hormones increase your risk of rhinosinusitis or anosmia?

Which viruses/infections put people at risk of developing anosmia?   What are the risk factors in the individual for developing allergic disease?

Causation:

What are the underlying cause or causes to develop preventative measures or treatments for anosmia (with or without taste disturbance) and allergy?

Triggers:

What is the role of household cleaners and synthetic perfumes in triggering sore throat or rhinitis? What are the dietary triggers for rhinitic symptoms?

Links to other conditions:

What is the link between allergic upper airways disease and lower airways disease, and what are the ways of reducing the risk of progressing to the latter?

What is the link between CRS with nasal polyposis and asthma; does surgical or medical treatment prevent progression to the latter?

Investigate the possible links between allergic rhinitis, thyroid function and autoimmunity overall.

Pathogenesis:

Better understand inflammatory pathways including TGF-α, T-cells, dendritic cells, microbiomes.

Understand the pathogenesis of 'allergic' fungal rhinosinusitis and AERD (aspirin-exacerbated respiratory disease). What is the role of adrenoreceptors in rhinology?

How can we use animal models to better understand sinonasal disease?

How conditions progress:

What is the prognosis of untreated smell and taste dysfunction, including impact on other health issues?

Conduct multicentre RCTs on surgery versus no treatment for patients with CRSwNP to establish the natural course of disease.

Indirect healthcare costs: What is the impact upon daily life, the economy and performance of the increasing incidence of allergies and allergic rhinitis?

Impact of "hidden" conditions: How does having a "hidden" condition, such as loss of sense of smell, loss of sense of taste or allergies increase frustration and suffering due to the impact on social interaction, family interaction, interaction with healthcare professionals and enjoyment of life?

Daily functioning and wellbeing:

What is the impact of loss of sense of taste and smell on eating a normal, healthy, safe diet? What is the impact of anosmia on eating habits and wellbeing of adults and children?

What is the general quality of life (no specified domains) for patients with anosmia, loss of sense of taste or allergy? What are the levels of distress/daily suffering caused by anosmia?

Psychosocial impact:

What is the impact of anosmia and loss of sense of taste on social isolation and withdrawal? What is the impact of anosmia and lack of sense of taste on confidence and self-esteem?

Relationships: What is the impact of anosmia on personal and intimate relationships?

Fear and safety issues: Understanding the patient experience of fear over safety (food, gas leaks, smoke) in anosmia.

Prevention of Ill Health, Maintenance of Good Health

Primary prevention:

Is there a role for dietary modification in the prevention of sinus disease and allergy?

Would controlling pollution levels have an impact on sinonasal and taste problems and general respiratory health? Are there strategies to prevent acute otitis media in children?

Does regular microsuction prevent otitis externa?

How can the use of simple interventions such as hand hygiene prevent ENT infections?

Secondary prevention: Are there risk factors for the progression of ARS to CRS and can this progression be stopped by active intervention?

Clinical Effectiveness

Diagnostic testing:

What is the role of diagnostic aspiration in acute otitis media?

How can we improve diagnostic radiological scanning techniques and interpretation for mastoiditis? Is there a need for standardised diagnostic testing to be developed for anosmia and taste dysfunction? Should the number of allergens tested for in skin prick tests be increased?

Diagnostic criteria:

What are the diagnostic criteria to differentiate tonsillitis from sore throats?

Develop diagnostic criteria to identify treatable causes of anosmia and taste disturbance.

Timeliness and accuracy of diagnosis: What strategies can we design for work-up of possible primary immune deficiency in patients with recurrent sinusitis as there is a recognised concern regarding diagnostic delay (average 10 years) of primary antibody deficiency that may present with recurrent sinopulmonary infections?

Existing therapies:

Recurrent tonsillitis:

•What is the cost-effectiveness of tonsillectomy?

•What is the role of tonsillectomy for severe recurrent sore throats?

•Does reducing the number of tonsillectomies performed have an impact on health in adulthood?

•Assess the impact of reduction in number of tonsillectomies on hospital admissions for tonsillitis and its complications.

•Which antibiotic should be used for acute episodes and how long should the course last?

Preventing complications of ENT infections:

•Does the early use of decongestants for 'head colds' prevent the onset of acute sinusitis for people who are prone to this complication?

•How can early management of infections be improved to prevent progression to perforation of the eardrum, mastoiditis, meningitis or severe tonsillitis requiring prolonged admission?

Can prophylactic antibiotics help prevent frequent ear infections? What combined interventions can be used in the elimination of biofilms?

Assessing antibiotic use:

•In upper respiratory tract infections/conditions.

•Are ciprofloxacin eardrops used excessively and how can we avoid resistance?

•Role, type, duration of antibiotics for AOM.

•Recurrent AOM and antibiotic sparing approaches: is there a role for grommets? What is the best management of suppurative otitis media in primary care?

Does a single annual steroid injection for hayfever have any significant side effects? What is the safest antihistamine to use in pregnancy?

What are the post-operative effects of using Moffet's solution in the nose prior to nasal surgery?

Whether doing a full FESS gives better long-term outcomes than a minimal FESS in CRSwNP and CRSsNP: would standardisation of the extent of endoscopic sinus surgery reduce complication rate?

What is the effectiveness of balloon sinuplasty?

How effective are routine surgical interventions in the nose: septoplasty, SMD, turbinoplasty?

How effective is sinus surgery in preventing further relapses of sinus conditions or does it exacerbate the problem? Are patients compliant with long-term macrolide therapy, and if so how many of them actually get their LFT's and ECGs monitored?

What is the effectiveness of antibiotic use in CRS? Are treatments for post-nasal drip effective?

Assessing the treatment of allergic and perennial rhinitis in primary care. What is the effectiveness of topical steroid drops in symptom control?

Addressing the lack of reliable data on effectiveness of saline douches/irrigation.

Conduct an RCT studying the effects of oral corticosteroids on olfactory function in CRSwNP.

What is the effectiveness of short course oral steroids in primary care on recurrence of nasal polyps?

The clinical and cost effectiveness of long-term macrolide use in CRSsNP: do they reduce the need for FESS, and if so, for how long?

Improve anosmia treatments to prevent risk to safety from spoiled food/smoke/gas leaks: can smell training benefit anosmia sufferers?

What is a good framework for management of atypical facial pain?

Improving therapies:

How can we improve the effectiveness of oral antihistamines? How do we improve outcomes for patients with anosmia/CRS?

Improvements for ESS: better pain relief post op, research use of local anaesthetics rather than general.

Comparing therapies:

What is clinical and cost-effectiveness of endoscopic sinus surgery with medical therapy versus medical therapy alone for CRS with nasal polyps?

What is the benefit of sinus surgery compared to medical treatment in chronic sinusitis?

Timings of interventions:

Does early surgical intervention chronic rhinosinusitis with nasal polyps alter the course of the disease? Does a delay in being treated reduce the possibility of a cure for anosmia?

Adopting treatments:

Immunotherapy:

•The role of sublingual and subcutaneous immunotherapy in the management of allergic rhinitis (seasonal and perennial).

•Can the NHS offer more effective treatments for hayfever sufferers, such as immunisation? What is the effectiveness of avastin in HHT?

Selecting patients: Which patients with allergic rhinitis should be referred for immunotherapy?

New therapies:

What can be done to eliminate ear infections in children/older people?

How can the sense of smell be improved or restored in patients with Kallmann's syndrome?

Seek better local therapies for immunomodulation.

What is the role of immunotherapy in chronic rhinosinusitis with polyps?

Develop and assess the use of Protopic for chronic otitis externa unresolved with regular drugs.

Is there potential for use of stem cells/tissue engineering or regeneration to restore sense of smell in anosmia?

Targeted therapy:

Is it possible to analyse local IgE production to target immunotherapy in hayfever?

Study with adequate numbers and ENT assessment to establish when cows milk protein intolerance causes or worsens nasal/sinus congestion: if there is a link are there any biological markers (possible role of component assays)?

How do we perform proper phenotyping of patients with CRS to guide treatment?

Identify subgroups to customise treatment:

Demonstrate whether the relative frequency of different symptoms and signs in CRSwNP and CRSsNP predict a differential response to different therapies.

Develop new classifications based on endotypes and phenotypes to determine treatment options for olfactory disorders, headache and facial pain.

Outcome measures:

How do we develop condition-specific quality of life outcome measures for rhinology? Develop PROMs in rhinology (septorhinoplasty/anosmia treatment).

Use consensus-based approaches to develop outcome measures, especially to determine what domains of impairment are important, for nasal surgery.

Designing and Delivering Healthcare

Patient pathways:

What is the unmet morbidity due to lack of referral from primary to secondary care for chronic rhinosinusitis? What is the impact of delay in referral on patient health, wellbeing and quality of life in anosmia?

How do we increase awareness of and streamline referrals to subspecialist taste and smell clinics? Standardising emergency pathways in ENT, especially in epistaxis.

Is there a need for a clear pathway for chronic rhinosinusitis/anosmia; what would the impact be of this pathway? How do you incorporate emotional and psychological support in the chronic rhinosinusitis/anosmia pathway?

Allergy clinics:

•What is the need for more specialised allergy clinics?

•What are the models for combined allergy clinics and how do you set them up?

•What is the impact of multidisciplinary care and nurse-led support in allergy clinics on QoL outcomes? Smell and taste clinics:

•Is there a need to increase the number of specialists and specialist clinics for smell and taste disorders?

•How do we increase awareness of existing smell and taste clinics in patients and clinicians?

•Should smell and taste clinics be established in Scotland? Is there a need for a pathway for atypical facial pain?

What is the clinical and cost-effectiveness of primary or community care for routine microsuction and treatment of otitis externa?

Improving service: Develop outcome measures that determine the quality of ENT care in primary care, such as whether rhinitis treatment is leading to better asthma control.

Barriers to access:

How does fear of examinations and procedures, especially with skull base conditions, impact on patients seeking help for sinonasal disease?

How do the attitudes of patients, GPs and ENT specialists to the treatments currently available for CRS sufferers affect access to service?

Practice variation and guidelines:

What is the adherence to EPOS guidelines in primary and secondary care? How can we improve implementation of EPOS guidelines?

Assessing and addressing variation in the management of ear infections in primary care. Expanding current guidelines for antibiotic use in upper respiratory tract infections.

How do we effectively minimise the antibiotic burden in children with upper respiratory disorders?

Communication

How can healthcare professionals improve listening and communication skills, be encouraged to acknowledge patients' symptoms and experience of disease, and address patient concerns about anosmia?

What is the impact on patient outcome of using pharmacists to give information and advice on using nasal sprays? How can we raise public awareness of the "hidden" condition of anosmia and its impact on psychosocial wellbeing and interactions? What is the impact of raising awareness on sufferers?

What is the impact of anosmia support groups on condition-specific and psychosocial outcomes?

What is the impact on care and outcome of making patients aware of new areas of research like smell training? How do we standardise consent for nasal polypectomy?

Dissemination of evidence: How much is known about the evidence for tonsillectomy for recurrent tonsillitis among non-ENT clinicians; how can we improve dissemination of this evidence?

Education

How much is known about otitis externa and its management in primary care? What is the impact on patient care and outcomes from training GPs in the management of otitis externa and eczema?

What is the impact on patient care and outcomes of teaching healthcare professionals about the effect of rhinosinusitis on quality of life, patient safety and health?

How can knowledge and management of rhinosinusitis be improved in primary care?

Sustainability of care

Understanding Health and Illness

Incidence and prevalence:

What is the prevalence of ENT conditions in primary care?

Using incidence and prevalence of disease to determine unmet need in the community and primary care.

Links to other conditions: What is the impact on society of ENT manifestations due to neurodegenerative disorders?

Prevention of Ill Health, Maintenance of Good Health

Secondary prevention:

Is an adult hearing screening programme cost effective?

What is the socioeconomic impact of a screening programme for age-related balance problems?

Tertiary prevention: What is the impact of developing rehabilitation programmes and self-help strategies for hearing loss and vestibular dysfunction on reducing need to utilise primary and secondary care?

Clinical Effectiveness

Existing therapies: What ENT surgery is most cost effective?

Outcome measures: How do we consider the effectiveness within the NHS of any improvements in technology, products or interventions?

Designing and Delivering Healthcare

Patient pathways:

What are the direct and indirect healthcare costs of establishing specialist clinics for "dizzy" patients?

How do we balance cost-effective pathways with best care in the ENT medical management of children and adults? How do we sharing and replicate models of good care nationally?

What is the evidence base for existing pathways?

How safe are virtual clinics and for which conditions are they suitable?

How do we develop online or mobile phone programmes for delivering treatments and interventions? What is the clinical and cost-effectiveness of community-based ENT emergency clinics?

What are the ways and cost-effectiveness of incorporating integrated natural health care in pathways?

How can waiting times for ENT services be reduced; is there a role for business models in assessing problems and delays in the referral process?

Treating patients in primary care and the community:

What ENT conditions can be safely and effectively managed in primary care or the community instead of secondary care?

What is the impact on waiting times and demand on acute care by providing these services here? Are services provided here cost effective?

How do we provide these services?

How do we increase the ability of GPs to treat ENT conditions in primary care?

The resource implications on primary care: how do we fund and support practices to provide this care?

Audiology-led services:

What is the impact of audiology-led triage for balance, dizziness and hearing loss issues on ENT capacity problems and waiting times?

What additional resources and support would be needed for audiology-led services? How could these services be made more accessible to people who work?

Hearing aids and services:

How often (and why?) patients find better provision through private dispensing?

What is financial implication of non-use of hearing aids that have been given to patients?

Current and future services:

How do we develop a comparative scale of importance based on value to society for ENT services/ procedures compared to other services/procedures?

Does ENT provide value as a specialty in state---delivered health care? Should this care be delivered outside the NHS? What is the impact of increased management in primary care on the sustainability of ENT services?

Modelling to predict future need for ENT services to determine resource needs for a sustainable service.

What factors and working conditions affect the health and wellbeing of ENT services' staff and how do these ensure sustainability of services?

Are current resource needs being met for ENT equipment in community, primary and secondary care?

Commissioning:

What is the value (direct and indirect) of commissioning common procedures? Where in ENT can we/should we restrict services?

How do we develop evidence to justify the commissioning of new technology or techniques?

Is there a benefit to a system of private or NHS contribution to commissioning hearing aids outside of the NHS? What is the effect of NHS privatisation and downgrading of NHS treatments that might be offered in future?

Communication

What strategies (IT, websites, helplines) would make information on causes and treatments of conditions accessible to patients? What impact would such strategies have on help-seeking behaviour?

What is the impact of raising public awareness of ENT conditions on the referral rate to ENT services?

What is the impact of efficient dissemination of evidence-based guidelines on outcomes and referral rates to secondary care?

Education

Training surgeons for the future:

How do we develop more efficient and effective methods of surgical training? What models can be used to establish and develop academic training in ENT? What is the extent and impact of bullying on ENT trainees?

Primary care:

How can tests and treatments be taught effectively to healthcare professionals in primary care? What impact does this have on referral rate to secondary care?

Establishing a Research Culture

Value in research:

Developing a "scale of importance" for different interventions both within ENT and with other specialties in order to determine which research provides most value to society.

Could we develop the equivalent of a QALY combined with disease prevalence/incidence and cost to society in ENT to rank the conditions to ascertain how much research funders' money should be spent?

Balance long---term high quality trials with small rapid projects that can be delivered in an integrated way throughout all the different areas of provision in healthcare.

Develop research in areas that provide maximum benefit (ie in areas where little is known or understood) to large populations.

How do we establish a culture of value in research where it is acceptable to fail, and to learn from failure?

Engagement and collaboration:

With whom do we need to engage and how do we put processes into place to rapidly implement good quality evidence? How can commissioners be engaged and encouraged to promote research alongside service commissioning? With whom do we need to engage and how do we put processes in place to facilitate the trialing of new devices and interventions?

Technology and research:

How do we design databases/electronic health records/registries that allow us to collect real world patient data and utilise it for research?

How can we utilise existing databases, like CPRD, to understand disease (track epidemiology, natural history and treatment modalities)?

Global perspective: What cost---effective models of healthcare from the developing world can be used in the UK?

Table 1 The long list of research ideas

287

Appendix 10: Expert Forum report

GENERATE Expert Forum Report

Expert Forum Report

Introduction

This report has been produced following the GENERATE Expert Forum that took place on 1st June, 2015. It is divided into four parts:

1. **Overview of GENERATE:**
   This section gives a brief overview of the background, methods and results of GENERATE, and an outline of the process for the Expert Forum.

2. **Changes and Further Steps Following Expert Forum:**
   This section summarises the changes to the Framework and topic areas following the Expert Forum, and the further steps that are being taken in analysis and preparation for the Consensus Conference.

3. **Values and Criteria for Prioritisation:**
   This section summarises the discussion around the values and criteria that the participants felt were important in deciding what are important topics for research. It combines the pre-meeting feedback that was received as well as the discussion from the meeting.

4. **Expert Forum Poster Notes and Discussion:**
   This section reproduces the notes made on the posters and summarises the discussions that were had about the topic areas both during the small group and the dinner plenary session. It also includes some of the post-meeting feedback that has been received to date. At the end of the section there is a summary of some of the non-topic specific discussions.

We value your feedback on this report. We want to ensure that it accurately reflects the discussions that were had, and we would also welcome your thoughts and any further suggestions on the questions included following your further reflection. Please contact Natalie Bohm (n.bohm@ucl.ac.uk) with your comments.

Thank you again for your participation and ongoing support of GENERATE.

Part One: Overview of GENERATE

Background

ENT-UK, the BSA and the BAA have recognised that the research base for ENT, Hearing and Balance care needs to be developed in order to generate the high quality evidence that underpins the clinical practice and service delivery. This development takes time and planning, recognising that competition for research resources is strong. GENERATE was developed to address this need.

GENERATE is a research priority-setting exercise leading to the development of a research agenda. The design of the exercise was based on a narrative review of similar national projects. It uses an emergent multi-stage, consensus-based democratic approach to develop an agenda that will focus efforts and coordinate research in ENT, Hearing and Balance care. The process is being overseen by independent Steering Committee composed of stakeholder representatives.

Methods

Initial stakeholder mapping was undertaken by the academic team in discussion with ENT UK, the British Academy of Audiology and the British Society of Audiology. The Steering Committee was formed from representatives of these stakeholders, and expansion of the stakeholder map was done in discussion with this Committee. Data for GENERATE were gathered through a survey and focus groups. A wide range of organisations based on the stakeholder map were contacted to distribute publicity on GENERATE and disseminate the survey. These included healthcare professional organisations, charities, aggregate organisations and Clinical Commissioning Groups. The GENERATE survey was publicised through online patient involvement websites, and was promoted at local and national clinical research awareness events to the general public as well as professional conferences. Respondents to the survey were asked which topics, issues or questions about ENT, Hearing and Balance care were the most important and why they were important.

Focus group participants started with these questions and then engaged in facilitated, focused discussion to explore these areas in more depth and identify emerging themes in the conversation. The discussions were facilitated by two moderators, and were recorded and independently transcribed.

The data were analysed using a framework analysis (Ritchie, Spencer *et al* 2003). This was by undertaken by two independent researchers, with the senior academics of the research team (Professors Fulop, Marshall and Schilder) each analysing samples of ten percent of the data. Each analyst identified themes in the data. The themes identified were discussed by the research team and a framework of seven themes was developed. Subthemes were identified by two researchers independently coding the data back into the framework, and these were compared and refined. After agreement on the framework the researchers coded the data back into the agreed themes. After coding into the framework was completed, cross-cutting topic areas were identified by two researchers independently. Throughout the process of developing the framework and the topics discussions between the researchers have resulted in slight changes in code or theme names to more accurately describe and reflect the data following the principle of "define and refine" (Braun & Clarke 2006).

This analysis resulted in a long list of questions under the framework headings in the different topic areas. In order to put these questions into the context of current evidence and ensure that they have not already been addressed by research, an Expert Forum was convened. The purpose of this was to reduce the long list of questions to a shorter list for prioritisation. The participants in this group were determined by suggestions from the Steering Committee, and both represented the stakeholder map and had expertise in the topic and framework areas. The meeting consisted of plenary and small group work, with discussion based on the following three questions:

- The values and criteria that participants have used to decide what are important topics to research.

- Participants' expert opinion on the need for more research in the areas that were suggested in the long list of topics, with consideration of the current state of evidence in these areas.

- What type of research would be of highest value and doable in those areas?

The meeting was facilitated by a professional independent facilitator, with two researchers observing and taking field notes. After consent, the discussions were recorded for later analysis. Participants were also requested to record their ideas and suggestions for the shorter list of questions on posters of the topic areas, and were asked to provide further feedback after reflection in the weeks following the meeting. The data from the Expert Forum were transcribed and independently analysed by the researchers to develop a list of criteria and values used by the experts in determining shorter lists, and to capture the shorter lists with the discussion about their formation and contents. These will be mapped back to the original data so that it is clear what has arisen from this data and to identify any new topics introduced by the experts. The results of this discussion and analysis will be presented to a Consensus Conference which will prioritise the shorter lists to form the research agenda.

Results

The survey had 685 respondents. Outside of the six key healthcare professional groups identified (Audiologists, Audiovestibular physicians, ENT surgeons, GPs, Nurses and Speech and Language Therapists), respondents identified themselves as belonging to 21 other professional groups. These included paediatricians, geriatricians, physiotherapists, clinical psychologists, pharmacists and teachers of the deaf. Non-healthcare professional respondents identified themselves as patients, patient carers, parents, members of the public, voluntary workers and charity organisations.

Five focus groups were run with the following groups (*43 participants)*:

- Mixed specialist clinicians (*n=9*)

- Industry representatives and clinical researchers (*n=16*)

- Patients (*n=10*)

- GPs, Community Pharmacists and Practice Nurses (*n=5*)

- Service commissioners and managers (*n=3*)

A framework of seven themes was developed.

The agreed themes derived from the data were as follows:

Prevention and public health
Designing and delivering healthcare
Communication
Education
Understanding health and illness
Clinical effectiveness
Establishing a research culture

Nine topic areas were identified:

- Adult Hearing Loss and Tinnitus

- The Ageing Population

- Balance Problems

- Children and Young People

- Head, Neck and Airway

- Mental health and dementia

- Patient-centred care

- Inflammation, Infection and Allergy

- Sustainability of care

There were 21 active participants in the Expert Forum. The Framework and topic areas were discussed in small groups and in an extended plenary discussion. The discussion and key ideas were captured by audio recordings and by interactive posters that the experts wrote on to highlight areas of importance from the small group discussions. The results of this and the discussion in greater detail are presented in the following three sections.

Part Two: Changes and Further Steps Following Expert Forum

1. <u>Changes to Framework</u>

Following the discussion about the title of "Education" and concerns that this will not resonate with funders, the academic team has proposed combining the Communication and Education themes into one theme that encompasses Knowledge Mobilisation and Behaviour Change. The academic team is reviewing the original data to more clearly refine this theme. Other minor revisions to the theme titles have been made in order to better reflect the original data and the feedback of our experts. The revised framework would be as follows (Note "Behaviour Change" is a provisional title only):

Understanding health, illness and disease

Prevention of ill health, maintenance of good health

Clinical and-cost effectiveness of diagnosis and treatment

Designing and delivering healthcare

Behaviour change

Building capacity and capability in research

2. The original nine topic areas have been reduced to six topic areas:

- Adult Hearing Loss and Tinnitus
- An Ageing Population
- Balance Problems
- Children and Young People
- Head, Neck and Airway
- Inflammation, Infection and Allergy

3. These changes, and the recommendations in the topic areas from the experts, will form the basis of the briefing to be sent out to the attendees of the Consensus Conference. The academic team is mapping the recommendations from the Expert Forum back to the original data to ensure that an appropriate spread of responses has been represented, without undue bias or weighting being given to any one group. It should be noted that there was extended discussion about conscious and unconscious bias by the experts in the forum, and the Preliminary Report from this meeting has been circulated to them following the meeting for further comment and consideration.

Part Three: Values and Criteria for Prioritisation

1. Factors based on patients/individuals:
   - Is the question important to/supported by patients?
   - What are the unmet needs of the patients (demonstrable unmet need to patients which can be a small but very disadvantaged patient population/research in areas where there is a well-established need for interventions and innovations)?
   - What is the value to patients (this could be a relatively small positive effect to many patients or a big effect for the more rare conditions)?
   - Will the research have an impact on individuals and their families?

2. Transformative/Aspirational change:
   - To research:
     - Will this change to way we look at research?
     - Will this introduce new ways of doing research to the field (different study design/new data sets)?
   - To practice and interventions:
     - Will this generate new, transformative knowledge?
     - Is this more than an iterative, incremental improvement?
     - Will research in this area have a wider impact on other topics (orphan conditions having wider impact/knowledge developed has other predictive indicators for a patient)?

3. Implementable improvements/pragmatic change:
   - To practice:
     - Is this an area of true equipoise between clinical choices (is this a true uncertainty/is this an area of wide practice variation)?
     - Would the change in practice be cost-effective (will the NHS implement it)?
     - Will this impact on patient care in a good time frame?
     - Will this change practice (are we bad at managing this/have we evaluated existing treatments)?
     - How can we harness the latest in therapeutic breakthroughs?
   - To service:
     - Will this improve efficiency of service (consider political drivers to reduce spend/AQP/rationing – answers that address this)?
     - Will this impact on quality of care and safety?
     - Is this an area where new treatment modalities and interventions in the organisation and delivery of care have been designed with service users in mind?

4. Factors based on conditions:
   - What is the cost of the disease/condition to healthcare (rare conditions can have large cost implications as well as common conditions)?

- What is the impact of the condition on society?
- Is it frequently occurring in clinical practice?
- What is the incidence and prevalence of the condition/size of the population affected?
- What is the importance of the health problem as represented by high burden of disease (as a combination of prevalence and impact on quality of life/ combination of frequency and resource burden)?

5. Practicalities of research:
   - Capacity:
     - Is the skill set to conduct the research available?
     - Can we utilise or maximise existing resources in a new way (existing data sets/using knowledge or evidence from other fields/preventing duplication)?
   - Feasibility:
     - Is the research achievable?
     - What is the cost of the proposed project?
     - What is the length of time required to do the research?
     - Is there internal competition in recruiting patients (similar studies; studies targeting similar groups)?
     - Does this research fit into an overall strategic agenda?
   - Fundability:
     - Is funding available?
     - Would funders be interested?
     - Does it fit into existing funding work streams?
     - Does it reflect national/political/industry priorities (should it?)?

Tensions noted in the group discussion

1. Feasibility of research v possibility for transformational change; pragmatism v aspiration

2. Common v orphan; numbers v severity; population v personal

3. Broad topics v specific detailed research questions

4. Political agendas (govt/industry) v neutral agendas

5. Use existing knowledge v develop new knowledge

Adult Hearing Loss and Tinnitus

Understanding Health
- The burden and impact of hearing loss at an individual, family and population level.
- Thromboembolic disease and sudden sensorineural hearing loss
- Molecular characterisation of familial hearing loss

Prevention of Ill Health, Maintenance of Good Health

- Screening for hearing loss and dementia

Clinical Effectiveness
- Hearing aids for mild-moderate hearing loss
- Optimal management for the patient with a vestibular schwannoma
- Alternative test batteries to test hearing beyond PTA
- Hearing aid strategies – which works best
- Tinnitus management strategies and hearing aid/combination devices

Designing and Delivering Healthcare
- Optimum "system" to identify and treat those who would most benefit from aids to audition and/or tinnitus "therapy"

Establishing a Research Culture
- Can we develop effective medical and biological treatments for S/N hearing loss

Discussion:

General points:

- The above list is a distillation of several pages and some points combine many questions into broader overviews. Not everything has been captured and there was concern that it could be too distilled.
- Distilling the list involved trying to think at the individual and population level, as well as challenging some of the preconceived ideas of what actually works, such as HAs. Capturing

"blue sky thinking" was also important, hence the inclusion of topics involving recent and exciting developments in molecular therapy. Part of the discussion was considering whether HAs could be considered old-fashioned and whether there were more creative ways of addressing the problem. It was pointed out that auditory implants existed for all types of hearing loss but that this involved an is intervention and the cost of this needed to be justified. Again it was pointed out that these assumptions needed to be challenged and that there should be consideration of drug therapies for hearing loss.

- Is the accepted evidence up to date? Practice in this area and HAs have changed a lot since some of the evidence was developed. In this topic as well as the others we need to consider how to ensure that the evidence stays relevant.
- It was questioned whether we understand primary prevention sufficiently, and this area was seen as a gap. It was also questioned if there was a gap between what we know and what we practice.

Specific points on poster:

- Prevention: Could we consider combining screening programmes – hearing loss and dementia?
- Designing healthcare: Optimal system – this considers designing a whole system that identifies and then manages those that would or may benefit from HAs/audition/tinnitus therapy.
- It was questioned why mild hearing loss had been captured. It was pointed out that this had been raised in the original data. It was noted that up to 50%-60% of HAs dispensed were not utilised for a wide range of reasons, therefore the question of whether we should dispense HAs for mild/mod hearing loss was relevant as they are costly if not utilised. The question of whether we needed to educate those around them to use them more effectively was raised, and it was also felt that this overlapped with the Ageing theme and design issues around them being "too fiddly" or if there was a lack of understanding of the importance of using them. Again greater understanding of the long-term benefit of HA use may be important to this section.

New Points raised:

- It was questioned if work around whether hearing loss is a predictor of Alzheimer's, and who develops hearing loss had been done and if this should be added.

An Ageing Population

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Understanding Health
- Impact of HL on mental health, QoL, ability to participate in society
- Impact of age-related conditions on QoL and family "burden"
- Multimorbidity
- Genetics and heredity
- Effect of loss of smell

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Prevention of Ill Health, Maintenance of Good Health

- Screening for HL in the elderly
- What is the best screening method for HL in the elderly
- Self care for balance
- Prevention of HL/ protection against noise in young people
- Use of technology in self screening/prevention

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Clinical Effectiveness
- Interventions for HL in ageing populations
- Why do people use their hearing aids? – not technically helpful - do they not hear how to use them
- Effectiveness of screening for hearing loss
- Impact of screening for hearing loss
- Intervention for balance problem in elderly
- Timing of interventions
- How do we restore hearing in elderly ➔ discovery science so we do not need to wear hearing aid

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Designing and Delivering Healthcare
- Balance pathways
- Dizzy clinics – link to falls clinics
- Access to services – travel/balance problems
- How would multidisciplinary assessments benefit

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Communication
- Involve patients in designing interventions/therapies and strategies
- Education/communication about issues of noise causing HL
- Behaviour change for risk factors
- Improve listening and communication skills
- Patient safety: raising public awareness of balance conditions

</div>

Discussion:

General Points:

- There was reflection on the phrase "THE" ageing population as it is not a single population; it is not homogenous and it is changing. People now live longer with disabilities and illness, however it was pointed out that there is nothing unusual about older people except amount of disease you find. It is rare that a person only gets to age with just hearing loss; rather we should be considering the impact of multi-morbidity and polypharmacy. It also needs to be recognised that there is heterogeneity in ageing, for example, the experience of the disadvantaged elderly is different.
- There needs to be thought around the nature and impact of the problem, and a recognition that the impact extends to whole families.
- One of the key points is considering how people become patients, and the fundamental question of how we think about older people and how they think about themselves. This is especially true around hearing loss – is this actually a normal part of ageing?

Specific Points:

- Is self-screening feasible for older people? Similarly, is self-management possible?
- Are HAs the only solution? This overlaps with the discussion in Adult Hearing Loss.
- Balance – have physios and OTs got balance sorted? There should be further discussion with these groups. Do we treat balance alone or in combination with other clinics/conditions?
- Education – there needs to be more education around the diagnosis of H/L and balance in primary care.
- No comment was made on the effect of loss of sense of smell in this area, although there was discussion around loss of smell as a predictor for neurodegeneration in the Inflammation, Infection and Allergy discussion.
- Research culture – there is a need to find and train people who want to research in this area.

Patient-Centred Care

Designing and Delivering Healthcare
- Timely diagnosis and referral – support GPs
- Patient pathways
- Emotional/psychological support
- Tinnitus/depression
- 'Cochrane' type review of referral pathways and clinical pathways

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Education
- Many issues have been answered in other fields – can be applied to ENT
- Multidisciplinary post grad education

Discussion:

General Points:

- What does high quality ENT, Hearing and Balance care look like – can we take models from elsewhere? Consider Cochrane type review of best service and pathways and then consider how to apply to these fields. There was detailed discussion about 303eneralizable knowledge in the Sustainability of Healthcare discussion that elaborates on these points.
- Education – does this sit within a research agenda? Are we considering types of teaching or impact on patient outcome? We should be considering more education for all members of the MDT, not just selecting out certain professional groups. There was further discussion about the suitability of Education an appropriate term for the theme, which is detailed under Further Discussion.

Specific Points:

- Although all areas are important we need to consider where we should start. Here it was felt that there should also be a focus on appropriate referrals and GP support, and that this was true through most of the framework.

Mentrl Health and Dementia

Overlap Dementia with Ageing; Mental Health ➔ incorporate with other topics/themes ➔ cut across

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Understanding Health
- HL and cognitive health
- What is the impact on adult hearing screening programmes on adults newly diagnosed with dementia?
- Early diagnosis of dementia in anosmia

Clinical Effectiveness
- Timely and accurate diagnosis

Designing and Delivering Healthcare
- Can we standardise pathways/practice variations
- Safe follow up

Discussion:

General Points:

- It was felt that this was not a stand-alone area but more of a cross-cutting theme. It was felt that the questions around dementia and the role of hearing loss in neurodegeneration should be moved to Ageing. It was noted that there are levers here to get research done as it has been identified as currently politically important.

Specific points:

- It was noted that depression in tinnitus/hearing loss is important, and that this should be captured in the appropriate topic.

Children and Young People

Common childhood ENT disease: OME (glue ear); rAOM (recurrent middle ear infections); recurrent tonsillitis; hearing loss (SNHL/sensorineural hearing loss for example); SDB (sleep disordered breathing); nasal obstruction and rhinorrhoea (chronic).

Understanding Health
- Understanding natural history and aetiology
- Impact of common childhood ENT disease upon development: ➜ *carry over to prevention*
    - Healthy kids
    - Complex needs
- Greater understanding of the impact of hearing loss on educational attainment and language development

Clinical Effectiveness
- Research to fully evaluate current interventions in common ENT illness, with greater understanding that the most appropriate intervention may be influenced by age and subsequently change through childhood

Designing and Delivering Healthcare
- Development of decision aids for clinicians and parents:
    - OME
    - SDB
    - Airway obstruction in neurological disease and other complex conditions

Education
- Development of aids for parents and clinicians

Communication
- Better dissemination of evidence/knowledge; better healthcare professionals

Establishing a Research Culture
- Development of clearly defined outcome measures for particular conditions e.g. CoS [*Core Outcome Set*]

Discussion:

General Points:

- This has been distilled to 18 generalisable themes but it is recognised that there is a risk of it becoming too generic. In order to do this the focus has been on common or high impact conditions and their management. These conditions have been defined at the start of the list

above (note: SNHL put in because of high educational impact though not necessarily a common condition).

- This area starts at the bottom of the evidence ladder when compared to, for example, cancer. This may be why there are so many questions around the management of common illnesses and little discovery science. There is a need to fully understand what we currently do; to fully evaluate our research and evidence for current interventions and management. The concern with this is are we even measuring the right things at the moment to make such an evaluation meaningful?
- Children with complex needs are equivalent to older patients with multi-morbidity – we need to get away from single condition focus.
- Children grow, change and develop. Treatment and interventions need to change through childhood to reflect this. Thus in developing evidence we need to consider the influence of the age of child to gain a clear understanding of relative benefit, and how this changes as a child develops.

Specific Points:

- We need a greater understanding of the impact of common conditions on childhood development and education, such as a greater understanding of the impact of hearing loss (and single-sided deafness) on education.
- We should also consider other interventions beyond HAs, such as classroom interventions.
- The development of clearly defined outcome measures – a core outcome set – is vital. We might have missed key differences because patients have not had input into outcome measures; we need to understand the impact of conditions on patients and their families to develop this. The advantage of clearly defining outcome measures is that every study should then be reproducible and we should be able to combine data/evidence.
- Development of decision aids. There are concerns that even where we have evidence it's not being used and we need to think about the process of delivering information to parents and clinicians and developing decision aids. This may also help them understand why decisions on management may change through childhood.
- We need to improve our understanding of the natural history of common childhood illnesses.

New Points Raised:

- We also need a greater understanding of acceptability of interventions e.g. HAs, nasal sprays. Thus we need to build acceptability into study design and consider the impact of interventions (do grommets stop kids from swimming? Does going in for regular HA tuning have an impact on children and families?).
- Other areas that could be considered are paediatric tracheostomy protocols and issues around airway obstruction.

Inflammation, Infection and Allergy

Key Areas:

Rhinosinusitis
Sore throats
Otitis media

Understanding Health
- Burden – epidemiology
- Natural history
- Defining prognosis/predictors of outcome

Prevention of Ill Health, Maintenance of Good Health
- Life style
- Socioeconomic
- Environmental v host

Clinical Effectiveness
- Defining disease
- Populations ➔ Targeting patient care e.g. antibiotics
- Cost effectiveness – old v new
- Outcomes development (· ➔ *link to designing and delivering healthcare*)

Designing and Delivering Healthcare
- Examining barriers to care
- Understanding variations in practice

Communication
- Improving pt/clinician dialogue
- Risks/benefits/uncertainties

Education
- Role of pharmacists in ENT care
- Patient/primary care – consultation/prescribing and referral behaviour

Establishing a Research Culture
- Good observational data
- Willingness to include and randomise between surgical and non-surgical interventions (requires equipoise)

Discussion:

General Points:

- Three areas emerged as important: otitis media, sore throat and rhinosinusitis. These should be considered across the framework areas. Allergy and anosmia were not included despite frequently being mentioned as it was felt that allergy research could be and was being better led by other groups elsewhere, and anosmia was viewed as a symptom not a condition.

Specific Points:

- There is a lot of existing information on the prevalence of rhinosinusitis. It would be worthwhile considering defining population for paths of care by endotypes and phenotypes. This also allows for targeted therapy, which is important with regard to appropriate antibiotic usage as well.
- Prevention – environmental v host includes a variety of factors including diet/dairy.
- Research Culture: There is a need for acceptance and development of non-RCT research approaches including observational studies, for example on variation. Cultural change around equipoise and acceptance of randomisation, especially by surgeons, is also needed. Consideration needs to be given to equipoise in primary and secondary care and where patients are actually treated. Developing frameworks for better data collection including routine measures odd disease categorisation, intervention and outcome will be key to understanding quality and effectiveness of different interventions and will benefit observational studies and research.
- Understanding variations in practice is important as it may give insight into barriers, identify areas where there is lack of agreement about best care, and allow development of strategies under or over provision of interventions.
- Communication – improve patient understanding as well as patient clinician dialogue and consider how do we convey concepts of benefit v risk.
- Education – the role of the pharmacist interesting in considering their role in modifying risk factors and reducing burden on primary care. Also education in primary care, especially in discouraging inappropriate prescribing of antibiotics.

New Points Raised:

- Prevention – should this be broadened beyond diet to e.g. vitamin and mineral supplements? There is a lack of potential questions in this area.

Head, Neck and Airway

Understanding Health
- Understanding globus – causes and burden
- Facial disfigurement
- OSA

Prevention of Ill Health, Maintenance of Good Health
- OSA
- Screening programme for HNC
- Look into 2w wait
- Tertiary prevention
- Self management – snoring

Clinical Effectiveness
- MDT working – clinical and cost effectiveness across ENT
- Voice reinstatement for laryngectomy – new technologies; voice banking
- Improving diagnostics for HNC + emerging technologies
- OSA
- Tracheostomy – protocols and equipment
- Functional outcome measures patient PROM – link with COMET
- Globus pathway
- Pharyngeal pouches – management of
- Innovative therapies for facial disfigurement

Designing and Delivering Healthcare
- Care pathways for voice and diagnostics
- Tracheostomy care pathway
- 2ww for cancer

Communication
- MDT working

Education
- OSA
- Vocal hygiene

Establishing a Research Culture
- Accessing BRC's/BRU's
- Seizing funding opportunities (e.g. I4I, Wellcome, TA)

Discussion:

General Points:

- This was a process of distillation and it was felt this section was reasonably complete.
- Most of Head and Neck cancer is covered by the NCRI. It makes sense not to include HNC in this agenda as there are already multiple ongoing trials and resources are concentrated on current trials. NCRI studies include service delivery, rehabilitation, functional issues, and quality of life studies. Most research in this area is clinical trials based.
- There is a lot of potential for new technologies for voice and swallow.

Specific Points:

- Tracheostomies are a key area – consider designing new tubes and care pathways with clear decannulation protocols.
- PROMs – COMET project is ongoing but will only come out with patient priorities, so this needs to go to next level to develop outcome measures for ENT. Functional outcome measures: there are enough QoL outcome measures in cancer – is this true of benign disease (e.g. capture the effect of snoring on intimate relationships)?
- What is globus and how do we deal with it? Definition, diagnosis and underlying pathologies are not clear, and there is a lot of variation for globus management.
- Healthcare pathways – globus and voice – need pathway with proper diagnosis before can label something as functional.
- Laryngectomy rehabilitation – should this be included?
- MDT – it is important to consider the value of the MDT.
- Could we improve research culture in Head and Neck? Although there is a good research culture in Head and Neck cancer there is still a need to improve capacity; a focus should be given to improving academic careers for SLT.
- Snoring and OSA feature prominently in this section. There is a clear public health issue around weight and lifestyle with regard to OSA. Clinical effectiveness around OSA very important as there is little high quality evidence and few RCTs.
- Pharyngeal pouches – it was noted that NICE has given approval to GI surgeons for awake laser division of pouches, although this was based on poor evidence – a small RCT with 20% perforation rate and 2 mortalities. It was suggested that the question be broadened to management of pharyngeal pouches.

New Points Raised:

- It was noted that there was nothing on singers and voice in prevention.
- Facial disfigurement due cancer and trauma had been raised with regard to understanding impact and it was suggested that considering innovative therapies in this area be added.
- Research culture – ENT needs greater presence in front of funders. Currently nothing ground-breaking coming is seen to be coming from ENT. Need lab space? It was noted that a lot of money was going to BRUs and BRCs it is was asked if we are are we working with them; are we embedded enough? ENT needs to consider seizing opportunities and funding.

Balance Problems

> **Understanding Health**
> - The impact of balance disorders on QoL, work, societal burden

> **Clinical Effectiveness**
> - Optimising symptom control + rehabilitation in patients with dizziness and imbalance

> **Designing and Delivering Healthcare**
> - Impact on outcomes from designated balance centres <u>OR</u>
> - Optimum design of a population based balance service

> **Education**
> - Primary care management of BPPV

<u>Discussion:</u>

General Points:

- This section is INCOMPLETE but is felt to be a section of real importance. The impact of locomotion and balance on individuals, families and society and the healthcare burden of older people falling are <u>substantial</u>.
- Think of population based services – who are the people who are and aren't patients? Are we meeting their need? Get rid of primary/secondary care divide.
- Multi-morbidity overlaps with ageing and balance.
- There is a research capacity issue in finding people interested and capable in this topic. Research culture in balance research needs development; although the Audiovestibular community is more advanced, more cross-disciplinary work is needed and more capacity.

Sustainability of Care

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Understanding Health
- Developing stronger methods, patient reported outcomes, better measures of burden of disease/disability
- Individual experience + epidemiology
- Better epidemiology + better use of routinely collected NHS data
- Understanding the dynamics of illness trajectories + multimorbidity + treatment burden

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Prevention of Ill Health, Maintenance of Good Health

- Identify primary prevention methods and topics (gap!)

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Clinical Effectiveness
- Better evidence →guideline production & dissemination →improved knowledge transfer

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Designing and Delivering Healthcare
- What is the appropriate balance between primary/community and secondary care for different ENT/Hearing & Balance conditions?
- What can be safely and effectively managed in primary care and have patient acceptability? And/or self-management?
- What is already known in other fields and can be transported in?

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Communication
- Identify strategies (…) + learn from other areas of experience.

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Education
- Educating researchers + clinicians to deliver/apply research + knowledge
- Developing clinical academic pathways to build research capacity
- Development of multidisciplinary teams (learning from other specialties e.g. cancer/mental health)

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↓ Capacity/Capability Building ↓

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Establishing a Research Culture
- Link/collaborate with health improvement /HSR research + development in other fields
- Link and collaborate across wide range of stakeholders (clinical and research)
- Looking to future-proof knowledge and practice → horizon-scanning

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<u>Discussion</u>

General Points:

3 key things:

1. There is much research that does not need to be done because it has been done elsewhere in other areas – this is a really important message. A great deal is known about how to set up and disseminate evidence, plus a great deal is known about knowledge transfer and about getting evidence back into practice. This 313eneralizable knowledge needs to be acknowledged, sought after and applied in order to avoid unnecessary duplication. ENT needs to break out of its knowledge silo and this needs to be recognised in the agenda.

2. Much more needs to be known about the experience of disease and disability in this area. The former is very under-researched and under-known. This is true in the context of multi-morbidity (major topic of interest at the moment) and true in the context of not just burden of disease/symptoms but also burden of treatment. There needs to be greater awareness of the work that people with disability get delegated by the healthcare system in terms of managing technology and treatment modalities.

3. The importance of capacity building. There needs to be a cadre of people who can do the research. Should not delegate tasks to others – need people in these fields committed to clinical work and to clinical and health services research.

- A key gap in primary prevention was noted.
- We are looking at complex healthcare processes in which there are few individual decisions. There are lots of distributed decisions. These processes are powerfully shaped by other institutional forces – you need to look at the nature of the processes that you are interested in because those processes shape the capacity of a field to deliver care AND they create patients. Patients don't exist in nature; they are fabricated by humans. You are turned into a patient by a healthcare system that creates epidemiological populations as well as individual patients.

General group discussion

- There were concerns about how personal bias was managed and a recognition that everyone has their own interests and their minds are drawn those areas. As part of the process and in order to reflect on the decisions made and recognise if bias had been introduced, participants wanted to come back to these lists to comment after seeing the end product and having time for reflection. It was also noted that a mapping process was needed from the original lists to the selected topics so that there is accountability and a trail of evidence. This will distinguish between questions that come from data and those which were raised from experts de novo.
- The need for collaboration was emphasised throughout the discussion.
- Inequalities play powerful role in shaping health and services. There are wider policy problems here that need to be taken into account. We deal with diverse populations and we need to be cognisant of that and its impact. Those communities have not specifically been targeted but understanding of prevalence in different communities issues around access have been raised.
- Consider moving from condition-based approach to consideration of the patient, looking at particular problems faced by particular patients. Areas to consider are the sensory needs of multi-morbid older patient, younger patients with complex needs and disadvantaged patients.
- "Education" – thought needs to be given to this title and it may need to be conceptualised differently. Education a poor title generally as the key is to understanding behaviours, not just educational interventions: people can be taught but without understanding barriers to implementation it won't change clinical behaviour.
- ENT/Audiology/AudioVestibular – these are small communities and every patient should be offered involvement in a trial/research.
- Should we focus on improving existing technology or should we be innovative and transformative? Or can we do both?

References

Braun, V. and Clarke, V. (2006) Using thematic analysis in psychology. *Qualitative Research in Psychology* **3** (2): 77-10.

Ritchie J, Spencer L and O'Connor W. (2003). Carrying out qualitative analysis. In Ritchie J. and Lewis J., eds. *Qualitative Research Practice* London: Sage Publications Ltd. Ch. 9.

Appendix 11: Consensus Conference participant briefing document

Towards a National Research Agenda for ENT, Hearing and Balance Care

GENERATE Consensus Conference

13.30 – 17.30, 25th September 2015

The Farr Institute, London

Contact:
Natalie Bohm: n.bohm@ucl.ac.uk
Zaynab Ismail: zaynab.ismail@ucl.ac.uk

GENERATE Consensus Conference

How to use this booklet:

Welcome to GENERATE, the UK research priority setting exercise for ENT, Hearing and Balance care. This booklet contains background information on this initiative, an outline of what we are asking you to do, and the ideas we are asking you to consider including in the final research agenda.

You do not need to read the whole booklet, although you are welcome to if you would find it useful or interesting.

In your covering email you will have been asked to join one of the following small groups and focus on the relevant sections in this booklet:

Group 1: Adult hearing loss and tinnitus
Group 2: An ageing population
Group 3: Balance problems
Group 4: Children and young people
Group 5: Head, neck and airway
Group 6: Inflammation, infection and allergy in adults

Please follow the guide below that will direct you to the relevant sections of the booklet for your group.

All participants:
Read Introduction and Overview (pages 4 - 14)

- Group 1: Read pages 15 - 16
- Group 2: Read pages 17 - 18
- Group 3: Read page 19
- Group 4: Read pages 20 - 21
- Group 5: Read pages 22 - 23
- Group 6: Read pages 24 - 25
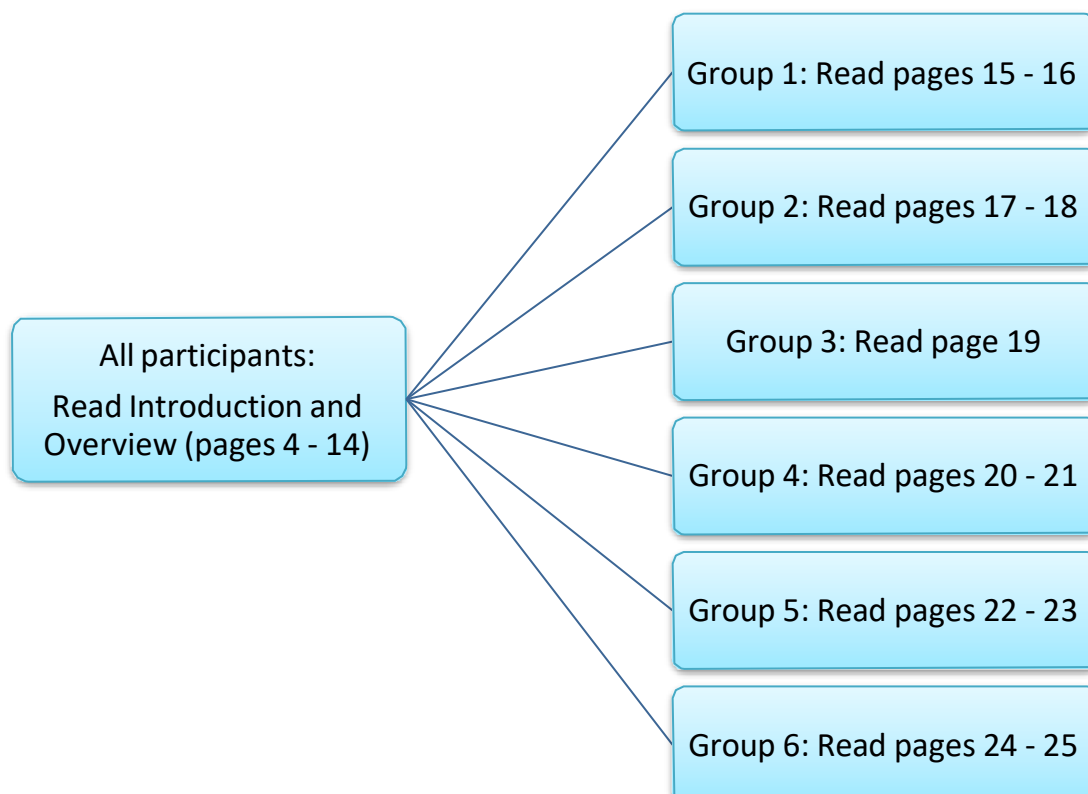
The following sections may be of interest or use to you:

If you would like an overview of all the research ideas being discussed at the GENERATE Consensus Conference please refer to Appendix 1 (pages 26 - 32).

There is a glossary of terms in Appendix 2 (pages 33 – 34) for your reference.

Appendix 3 (pages 35 - 36) is a list of the delegates attending the Consensus Conference.

Appendix 4 (pages 37 - 38) introduces you to the GENERATE team.

A detailed briefing document on GENERATE is available if you feel that more information would be useful to you. If you would like a paper copy of this document, or if you have any questions please contact:

Natalie Bohm: n.bohm@ucl.ac.uk for any questions on the content of this document
Zaynab Ismail: zaynab.ismail@ucl.ac.uk for any other questions.

Thank you for participating in the GENERATE Consensus Conference.

Consensus Conference: Preparation and Format for the Day

GENERATE has been initiated to develop a research agenda for ENT, Hearing and Balance care in the UK. It is a groundbreaking project that has been shaped by the views of patients, carers, healthcare professionals, researchers and other people that develop, use and benefit from evidence for clinical care.

We have gathered a list of research ideas that these people think should be considered for the final research agenda. The task for the Consensus Conference participants will be to decide on the ideas that should form the basis for the research agenda for ENT, Hearing and Balance care. We will not be trying to draft formal research questions or the full agenda on the day.

In order to decide the research agenda we will be asking you:

1. **To discuss what you think are the most important research ideas to be included in the agenda**
2. **Why you think these are important**
3. **To come to an agreement as a group at the end of the Conference as to what should be included in the final research agenda.**

These discussions will take place both in a large group and in smaller groups to discuss the specific topic areas in more depth (see Figure 1 below).

Conference starts: session with all participants facilitated by Phil Hadridge.

Small group sessions: facilitated sessions to choose topics in each group. Refreshment break provided.

Conference closes: Session with all participants facilitated by Phil Hadridge.
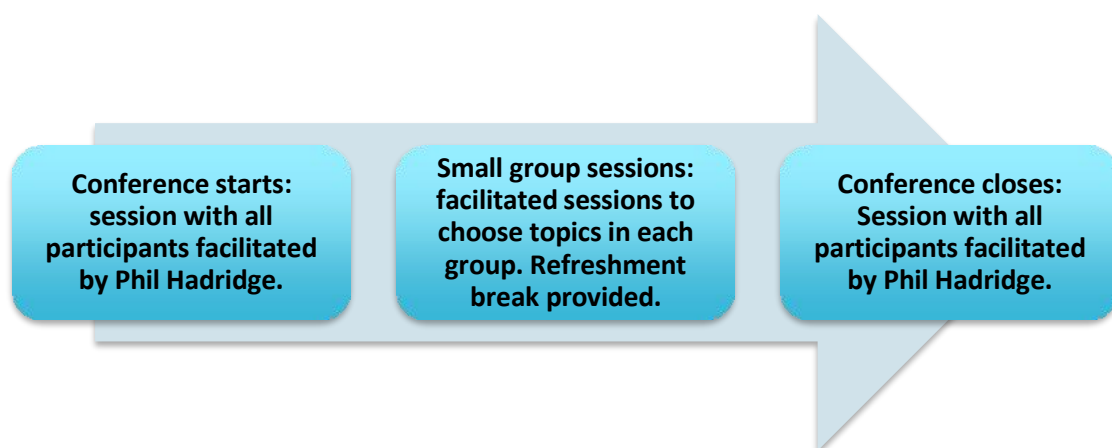
Figure 1: Consensus Conference: Structure of the day

We would like you to look at the ideas in your topic in more detail ahead of the conference, to consider which ones you feel should form part of the research agenda and why they should be included. Please choose two ideas as your top choices and two more as your second choices. You will be asked to feedback on this in your small group sessions.

Overview of GENERATE

Good clinical care makes use of the evidence developed through research; this tells us what works, what provides value, and how we should deliver our care. ENT-UK, the professional organisation of ENT surgeons, has recognised that the research base for ENT, Hearing and Balance care needs to be further developed in order to continue to provide the best care possible for our patients. This development takes time and planning, recognising that competition for research resources is strong. GENERATE was commissioned by ENT-UK, with the support of the British Society of Audiology and the British Academy of Audiology, to address this need.

GENERATE is a research priority-setting exercise that will result in the development of an agenda for future research. The process is being overseen by independent Steering Committee composed of representatives of the different groups that develop, use or benefit from evidence in this field.

An academic team (see Appendix 4), based at University College London, has designed GENERATE and is analysing the data. Data for GENERATE were gathered through a survey and focus groups. Respondents to the survey were asked which topics in ENT, Hearing and Balance care were the most important and why they were important. Focus group participants started with these questions and then had discussions to explore these areas in more depth.

There were almost 700 responses to the survey. Combined with the information from 5 focus groups, this meant that we gathered an enormous volume of information. Many of the issues raised fell outside of traditional clinical research that looks at the effectiveness of treatments. Yet these were the issues that our respondents felt were important. Instead of discarding the responses that fell outside "traditional effectiveness research," we decided to find a way to incorporate them in our agenda. To do this we used a technique called "framework analysis" (Ritchie, Spencer *et al* 2003[1]). The academic team used the data to develop a "framework" of themes that divided the responses into different aspects of

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[1] Ritchie J, Spencer L and O'Connor W. (2003). Carrying out qualitative analysis. In Ritchie J. and Lewis J., eds. *Qualitative Research Practice* London: Sage Publications Ltd. Ch. 9.

research. Some of the themes incorporate several different types of research that are linked by similar ideas. All the responses were entered into the framework, which still resulted in a large number of research ideas. Looking across the framework we found that we could divide the ideas even further by identifying common topics likely to be more familiar to our participants than some of the new ideas from the framework. These are the 6 topics that form the different small groups for this Conference.

After this analysis we still had a long list of ideas. This needed to be reduced to get a manageable number to discuss in the Consensus Conference. To do this we asked a group of experts, including patient representatives, to help us narrow the long list down. Based on their suggestions, and on the original data, we have developed the list of ideas contained in this booklet.

It is important to realise that the ideas in this booklet are not technical "Research Questions." Most of what we have included are broader areas to consider as priorities for research. This approach was truer to the data we got from our respondents and again we have chosen to incorporate this rather than dismiss what people have told us. Where possible formal research questions will be developed after the GENERATE agenda is agreed.

The Framework Themes

A framework of seven themes was developed and is displayed in Figure 2 below.

Figure 2: The GENERATE Framework

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1.   Understanding health, illness and disease

<u>Developing an understanding of ENT, Hearing and Balance conditions, and the impact they have on health and illness in individuals, their families and society.</u>

"...actually are we at the stage of talking about treatments? Shouldn't we focus more on aetiology, causation and these are areas that may not come up…" (GENERATE researcher focus group participant).

"I think maybe instead you could do some human research instead of some laboratory research, it would be good…" (GENERATE patient focus group participant).

2. Prevention of ill health and maintenance of good health

Strategies and interventions that prevent, stop or slow loss of function, and that maintain health and wellbeing among the population.

"Prevention is ALWAYS better than cure" (GENERATE survey respondent).

3. Clinical and cost-effectiveness of diagnosis and treatment

Testing existing and new ways to diagnose, treat and manage patients and their "conditions."

"In terms of the hospitals, nobody's asking me what works for me. Nobody's asked me about it or said what does work, what have you found to work for you? All of that information from all of these patients is waiting." (GENERATE patient focus group participant).

4. Designing and delivering healthcare

Strategies and interventions for improving the quality and value of health care, and making it sustainable and patient-centred.

"…there are so many areas, not only from the technological side but also from beyond the technological side how to develop the services, how to run the services which can be looked into purely from a research point of view. But it doesn't get discussed very often that is the problem." (GENERATE mixed specialists focus group participant).

6. Creating effective communication

Improving health and services through better communication and interpersonal interactions

"I would like to see consultants, GPs and other Allied Health workers show a greater understanding of the patients loss, fears, concerns and difficulties" (GENERATE survey respondent).

5. Promoting knowledge and learning

Interventions and strategies to improve health, function and service through knowledge sharing and learning strategies for patients, healthcare professionals and providers.

"… so because somebody else has taken the trouble to do the learning, she was able to share with me, no, you absolutely cannot even go in the water and do water aerobics, but because of her studies I'm able to do Tai Chi. Now you've no idea what that's done to my life. So I would like to see more people have a share in personal responsibility for their own life, because we have to accept that there isn't a cure for everything..." (GENERATE patient focus group participant).

7. Developing a research culture: capacity, capability and collaborations

Different ways of conducting research and embedding it in healthcare: value, capacity, collaborations, "real world data" and different ideas.

The issues raised in this theme of the framework were not researchable questions, but rather strategic ideas about how to make the GENERATE agenda successful, innovative and implementable. For this reason it is shown as a background to the other themes in the framework. It gives strategic directions about how to support and develop the research in

the other themes. The ideas raised here will form a strategy section in the final agenda and may be used to develop research questions.

<div align="center">**Topic Areas**</div>

Initially nine topic areas were identified. After discussion at the Expert Forum the academic team reduced these to six. The other three topics (Mental Health and Dementia, Patient-Centred Care and Sustainability of Care) were felt to be so important that they applied to all the topics, so their questions have been incorporated into the other six.

Adult Hearing Loss and Tinnitus

"…In silence tinnitus is always there to remind you something's wrong, and in noisy environments the deafness is always there to remind you something is wrong, and that's why it's incredibly insidious" (GENERATE patient focus group participant).

GENERATE respondents overwhelmingly felt that more research was needed in all aspects of hearing loss and tinnitus and in all age groups. Hearing loss and tinnitus in children and older adults is dealt with under the relevant topics, although there is some overlap between these. GENERATE respondents also recognised the need for more tinnitus research, and were clear about the suffering that people with this condition experienced.

An Ageing Population

"Well I think people are waking up to the big ticket items which is we're all getting older and, as we are fitter until we get older, that's only postponing the avalanche and people's expectations are still going up…" (GENERATE commissioner focus group participant).

GENERATE respondents recognised that an ageing population faced significant health challenges, often had multiple problems, and that ENT, Hearing and Balance issues could have a significant impact on them, their families and healthcare costs to society.

Balance Problems

"Balance disorders can have a catastrophic effect on the sufferer physically; emotionally; socially and in work settings. So continuing the promising research into causes and effective treatment are vital." (GENERATE survey respondent)

GENERATE respondents raised many questions about conditions affecting balance in all age groups. Our respondents felt it was under-researched and poorly understood, despite its significant impact on quality of life and ability to function on a daily basis. As one survey respondent noted when answering why this should be a priority: "You wouldn't ask if you had it."

Children and Young People

"These are all very common conditions of childhood which have a significant impact on the child a short and long term as well as their educational attainment." (GENERATE survey respondent).

GENERATE respondents were concerned about the impact of ENT, hearing and balance conditions on children and young people, who are "…yet to live their life" (GENERATE survey respondent). Children at all ages were considered, as well as children with complex needs, who often have ENT problems.

Head, Neck and Airway

"Massive burden of disease/illness in patients that receive no useful diagnosis or treatment from primary or secondary care. Massive expenses to the NHS in repeat appointments and recurrent investigations, without a conclusion every being reached" (GENERATE survey respondent, talking about globus pharyngeus).

This section deals with ENT conditions that affect the voice, swallowing and breathing. Many of the GENERATE respondents raised questions about Head and Neck cancer. Cancer researchers are already prioritising and developing ways of answering these questions. There is however a significant amount of benign disease that our respondents would like to

prioritise. This increasingly accounts for time taken in clinic, and many clinicians are uncertain about the best ways of managing these conditions.

Inflammation, Infection and Allergy in Adults

"It's not life threatening. It's not visible. However smell and taste are TWO of the senses. This has a huge effect on the quality of life on a day to day, hour by hour, minute to minute basis" (GENERATE survey respondent).

This topic includes questions about inflammation and infections of the ear, nose and throat, as well as allergic conditions affecting the nose and sinuses. Many respondents noted that problems with allergy were increasing, and a lot of questions were asked in this area. There is already considerable research happening, and our experts noted that some of these questions were being dealt with by immunologists and other specialists. ENT infections are very common, both in primary and secondary care, and affect both adults and children. Some of the questions raised in this section overlap with those in Children and Young People, and these questions have been placed in the topics where they appear most appropriate.

Values and Criteria for Prioritisation: Why are these questions important?

It is important to consider why the questions we choose are the priorities for research. One of the guiding principles of GENERATE is that it should be transparent, both in its process and in its decision-making. This is to ensure that no one group has undue influence or is biasing the final agenda. Part of this transparency is to understand the reasons for choosing the questions for the agenda, and this has been explored and discussed all the way through GENERATE. These ideas are summarised in Table 1 below. Some of the ideas discussed may seem to contradict each other. Should we choose pragmatic questions, or should we aim for aspirational projects? Do we pick topics that affect large numbers of people, or topics that have a major impact on only a few? Should we find better ways to use the knowledge we already have, or do we develop entirely new ideas? These questions do not have clear solutions, and we are not looking for right or wrong answers. It is more important that the

options are considered and weighed up so that we can reach consensus, even if we are not in perfect agreement with each other.

When deciding what questions you think should form part of the agenda, please consider these reasons and use them to justify your choices. If there are other reasons that you are using to make your decision then please make a note of them and they will be discussed at the Consensus Conference.

Factors based on patients/ individuals	- Is the question important to, or supported by, patients? - What are the unmet needs of the patients? This unmet need may be small but may affect very disadvantaged patients. Alternatively we may consider research in areas where there is a well-established need for interventions and innovations. - What is the value to patients (this could be a relatively small positive effect to many patients or a big effect for the more rare conditions)? - Will the research have an impact on individuals and their families?
Transformative/ Aspirational change	- To research: Will this change the way we look at research? Will this introduce new ways of doing research to the field, such as different study design or new data sets? - To practice and interventions: Will this generate new, transformative knowledge? Is this more than an iterative, incremental improvement? Will research in this area be generalisable to other conditions, especially when considering research into rare conditions?
Implementable improvements/ pragmatic change	- To practice: Is this an area of true uncertainty to clinicians? Is this an area of wide practice variation and will research change practice? Would the change in practice be cost-effective (and will the NHS implement it)? Will this impact on patient care in a good time frame? Will this let us harness the latest in therapeutic breakthroughs? - To service:

	Will this improve efficiency of service and influence spending or rationing? Will this impact on quality of care and safety? Is this an area where new treatment modalities and interventions in the organisation and delivery of care have been designed with service users in mind?
Factors based on conditions	• What is the cost of the disease or condition to healthcare (rare conditions can have large cost implications as well as common conditions)? • What is the impact of the condition on society? • What is the incidence and prevalence of the condition or size of the population affected? Is it frequent in clinical practice? • What is the importance of the health problem as represented by high burden of disease, as a combination of prevalence, impact on quality of life and resource burden?
Practicalities of research	• Capacity: Is the skill set to conduct the research available? Do we need to develop these ourselves, or do we need to collaborate with other disciplines? Can we utilise or maximise existing resources in a new way (existing data sets/using knowledge or evidence from other fields/preventing duplication)? • Feasibility: Is the research achievable? What is the cost and length of time required for the proposed project? Are other studies trying to recruit the same patients? • Fundability: Is funding available? Would funders be interested? Does it fit into existing funding work streams? Does it reflect national/political/industry priorities? Does this research fit into an overall strategic agenda?

Table 1: GENERATE Values and Criteria for Prioritisation

Group 1: Adult Hearing Loss and Tinnitus

Understanding Health, Illness and Disease

What is the burden and impact of hearing loss at an individual, family and population level? This includes the impact on quality of life, daily living, mental and cognitive health, and the impact on families and society.

What is the molecular characterisation of hereditary hearing loss? How can we use this to target treatment to individual patients?

Prevention of Ill Health, Maintenance of Good Health

How do we prevent hearing loss and tinnitus from developing? What is the risk of personal listening devices and environmental noise?

How can we prevent hearing loss developing from ototoxic drugs?

Clinical and Cost-Effectiveness

What is the clinical and cost-effectiveness of hearing aids for mild-moderate hearing loss?

What are effective rehabilitation strategies for single-sided deafness?

What is the optimal management strategy for the patient with a vestibular schwannoma/acoustic neuroma?

What are the best management strategies for tinnitus?

What is the evidence for auditory implant devices? For example, what are the benefits of cochlear implants compared to middle ear implants in patients with severe (not profound) hearing loss?

Which hearing aid strategies work best? Consider early intervention, user-friendly devices to improve adherence, and the role of emerging communication technology in hearing (such as digital communication devices, technology for self-fitting of hearing aids or brain-computer interfaces for controlling cochlear implants and hearing aids).

What alternative assessments should be used to test hearing beyond conventional Pure Tone Audiograms (PTAs)?

Can we develop effective medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?

Designing and Delivering Healthcare
What is the optimum "system" to identify and treat those who would most benefit from hearing aids or aids to audition? What is the optimum patient pathway for tinnitus?
Creating effective communication
How can healthcare professionals improve their understanding and communication with patients with tinnitus? What is the impact of individual and group support networks on patient outcomes for tinnitus and hearing loss? Outcomes include psychosocial and condition-specific outcomes.
Promoting Knowledge and Learning
Would raising public awareness of the "hidden" conditions of hearing impairment and tinnitus improve the quality of life and wellbeing of people with these conditions?

Group 2: An Ageing Population

<table>
<tr><td align="center">Understanding Health, Illness and Disease</td></tr>
<tr><td>

What is the prevalence and socioeconomic impact of hearing loss in the elderly in the UK?

What is the prevalence and socioeconomic impact of balance problems in the elderly in the UK?

What is the impact of hearing loss on older people with regard to mental and cognitive health, quality of life and the ability to participate in society?

What is the burden on family, carers and society of dizziness and falls in the older adult?

What are the mechanisms behind the link between hearing loss in the elderly and dementia in the elderly?

How can we identify and target patients at risk of age-related hearing loss?

Is loss of sensory function (hearing, smell) a predictor of age-related neurodegenerative diseases or Alzheimer's?

</td></tr>
<tr><td align="center">Prevention of Ill Health, Maintenance of Good Health</td></tr>
<tr><td>

Developing an adult hearing screening programme: consider practical and cost-effective methods for this, its impact on older people maintaining good health and function, and the feasibility of combining it with a dementia screening programme.

</td></tr>
<tr><td align="center">Clinical and Cost-Effectiveness</td></tr>
<tr><td>

What are effective interventions for hearing loss in an ageing population?

How can we restore hearing in older people so that hearing aids are not needed?

What are effective interventions for age-related balance problems in an ageing population?

What is the most effective way of assessing and managing the ENT manifestations, such as loss of smell, voice, swallow, of age-related neurodegenerative diseases?

</td></tr>
</table>

Designing and Delivering Healthcare
What is the optimum design for "balance pathways" (including vestibular rehabilitation) in older adults? How can GPs and hospital specialists best manage ENT conditions in older people with multiple medical problems? How can we improve access to hearing services for older adults who are housebound or in residential care, and for those with dementia? What can be safely and effectively managed in primary care, or by patients themselves, and would this be acceptable to patients?
Promoting knowledge and learning
What is the impact of raising public and professional awareness of hearing and balance problems as more than just a condition associated with age and infirmity? Consider the impact on health behaviours of patients, clinical behaviours of professionals and how they relate to long-term patient outcomes.

Group 3: Balance Problems

Understanding Health, Illness and Disease

What is the burden and impact of balance disorders at an individual and population level? This includes the socioeconomic impact on ability to participate in work to the individual and society.

What are the causes and course of balance problems, in particular Meniere's Disease?

How do we design, develop and utilise databases that capture "real world data", such as registries or electronic health records, to help us understand and manage ENT, hearing and balance conditions?

Clinical and Cost-Effectiveness

Develop clear diagnostic criteria and more sensitive tests, including cognitive processing tests, for the diagnosis of vertigo and balance disorders, including Meniere's Disease and vestibular migraine.

What is the role of diet and pharmaceutical drugs in restoring quality of life and independence for patients with dizziness and imbalance?

What is the role and impact of vestibular rehabilitation, including vestibular exercises, on the quality of life, mental health and wellbeing and independence of patients with dizziness and imbalance? Which patients benefit and what are the indications for referral?

Designing and Delivering Healthcare

What is already known about designing and delivering healthcare from other fields, and can this be adapted for ENT, Hearing and Balance care?

What is the optimum pathway or population-based service for the patient with balance problems? Is there a role for "dizzy clinics," Which specialties should be involved in them and where should they be based?

What is the impact of long waiting lists to access services on quality of life and mental health for patients with long-term vestibular and balance problems?

Promoting Knowledge and Learning

What is the impact on patient care, safety and referral rate of training GPs in the management of vertigo, and what is the best way of providing this training?

What is the impact on patient safety of raising public awareness of balance conditions?

Group 4: Children and Young People

Understanding Health, Illness and Disease

What is the cause and course of childhood ENT conditions? The conditions stated are
- glue ear,
- recurrent infections: middle ear infections, tonsillitis, rhinosinusitis,
- sensorineural hearing loss,
- sleep disordered breathing and
- nasal obstruction and allergies.

What is the impact of these childhood ENT conditions on the development and educational achievement of children, and on their families and society? What is the impact on otherwise healthy children and in those with complex needs?

What is the prevalence of paediatric balance problems and their impact on learning and locomotor ability? Can the impact be lessened if they are identified early?

What is the prevalence of paediatric tinnitus in different age groups?

What is the prevalence of unmanaged hearing loss in children immigrating to the UK and how should this be managed?

Clinical and Cost-Effectiveness

What is the best way to evaluate and manage children with tinnitus?

What is the best way to evaluate and manage children with balance disorders?

Develop clearly defined and agreed outcome measures to develop core outcome sets for childhood ENT conditions.

What are the effects of the use (and overuse) of antibiotics in children with recurrent ENT infections? What is the short and long term impact of reducing surgical rates in these children? What is the best management of children with recurrent middle ear infections and is there a role for diagnostic aspiration of middle ear fluid?

What is the role of grommets vs. hearing aids in otherwise healthy children with hearing loss due to glue ear, and in those with complex needs?

What is the clinical and cost-effectiveness of current interventions in childhood hearing loss: the effectiveness and tolerance of hearing aids in children; the effectiveness of classroom acoustic interventions; candidature for cochlear implants; multidisciplinary assessment tools for functional outcomes for hearing rehabilitation?

What is the best management strategy for children with allergic rhinitis? What is the role of immunotherapy?

How do we standardise the evaluation and diagnostic criteria for children with symptoms of obstructive sleep apnoea? What is the best management of these children, and how do we measure that?

Designing and Delivering Healthcare
What age-appropriate pathways can be developed for different types of hearing impairment that reflect the needs and development of children with and without complex needs? What is the appropriate balance between primary/community and secondary care for different ENT/Hearing & Balance conditions?
Creating Effective Communication
Developing and assessing shared decision-making tools for parents and clinicians.
Promoting Knowledge and Learning
How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professional in different settings?

Group 5: Head, Neck and Airway

Understanding Health, Illness and Disease

What are the causes and burden of disease of globus pharyngeus?

What are the causes obstructive sleep apnoea and its impact on the health, safety and employment of patients??

What is the impact of facial disfigurement, especially facial nerve palsy, on social isolation and withdrawal?

Prevention of Ill Health, Maintenance of Good Health

Can self-management strategies be developed and used to improve OSA and snoring?

Clinical and Cost-Effectiveness

How can speech rehabilitation or restoration post laryngectomy be improved, especially for women? How can advances in technology and materials science be utilised here?

Stapling of pharyngeal pouches: what is the long-term outcome? Have we wrongly moved away from open surgery, particularly in younger patients?

How can we improve the clinical care and safety of patients with tracheostomies both in hospital and in the community? Consider decannulation protocols and tracheostomy tube design.

What is the role for improved diagnostics for endoscopy (e.g. confocal imaging, contact endoscopy, narrow band) in head and neck cancer, and how can emerging diagnostic technology be used in the screening and management of head and neck cancer?

What innovative therapies for facial nerve palsies are clinically effective?

What is the optimal management for adults with OSA? Consider diagnosis, treatment, and its impact on general health.

What is the optimal management (investigation and treatment) protocol for globus pharyngeus?

What is the optimal management for adults with voice problems, including the role of voice rest, diet and pharmaceutical drugs?

Develop clearly defined and agreed outcome measures to develop core outcome sets for benign head, neck and airway conditions.

Designing and Delivering Healthcare
Develop a patient pathway for people with voice problems.   Develop a tracheostomy care pathway across hospital and community settings.

Creating effective communication
Assess communication in multidisciplinary care teams and its impact on quality of care.

Promoting Knowledge and Learning
What is the impact of raising public and professional awareness of OSA? Consider the impact on health behaviours of patients, clinical behaviours of professionals and how they relate to long-term patient outcomes.

Group 6: Infection, Inflammation and Allergy in Adults

Understanding Health, Illness and Disease

Define the different variations of disease that make up rhinosinusitis ("endotypes and phenotypes") and develop different ways of classifying them based on signs, symptoms and biochemical markers in order to target them with effective therapy and management.

What are the causes and course of chronic ENT infections in adults with and without treatment, and can we determine what predicts their outcomes? Consider risk factors and improving understanding of inflammatory pathways.

What is the impact on quality of life, relationships, mental health and wellbeing and daily functioning of the loss of sense and smell and taste?

What are the roles of chronic nasal conditions and their management on asthma and lower airways disease?

Prevention of Ill Health, Maintenance of Good Health

What lifestyle or environmental strategies can be used to prevent the development of chronic ENT infections and inflammatory conditions, such as the role of diet, supplements and pollution?

Clinical and Cost-Effectiveness

What are the diagnostic criteria for different subtypes of rhinosinusitis and can these be used to target specific therapies and identify "treatable causes?"

What are the most clinical and cost effective treatments for chronic rhinosinusitis? What is the optimal medical treatment (antibiotics, topical therapies, oral steroids in primary care, immunotherapy), surgical treatments (timing and extent and techniques of surgery, including balloon sinuplasty), or combination of the two for different subtypes?

What is the role for septoplasty or septorhinoplasty in patients with blocked noses?

Develop clearly defined and agreed outcome measures in primary and secondary care to develop core outcome sets for chronic ENT infections, inflammatory conditions and nasal surgery.

Designing and Delivering Healthcare
What are the barriers to accessing services for patients with smell and taste disorders? What is the impact of these barriers and delays in referral on these patients? What is the best pathway for patients with nasal allergies, and do we need to improve access to immunotherapy services? What is the role of multidisciplinary allergy clinics? How do we implement guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden in adults and children? What are the variations in adherence management guidelines for rhinosinusitis (EPOS guidelines) in primary and secondary care, and how do we improve their implementation? What is the variation in management of chronic ear discharge in primary and secondary care, and how do we address this?

Creating effective communication
How can healthcare professionals improve their understanding of and communication with patients with smell and taste disorders? What is the impact of anosmia support groups on the health and wellbeing of patients? How do we standardise consent for nasal procedures such as polypectomy?

Promoting Knowledge and Learning
What is the role of pharmacists in promoting understanding and giving advice about using medications for ENT infections and inflammatory conditions, especially topical nasal sprays? How can we raise public and professional awareness of the "hidden" problem of anosmia? Consider the impact on health behaviours of patients, clinical behaviours of professionals and how they relate to long-term patient outcomes.

Appendix 1: Broad Questions Across the Framework

This is a list of all the questions from the topics in the previous pages.

1.   Understanding health, illness and disease

How do we design, develop and utilise databases that capture "real world data", such as registries or electronic health records, to help us understand and manage ENT, hearing and balance conditions?

What is the burden and impact of hearing loss at an individual, family and population level? This includes the impact on quality of life, daily living, mental and cognitive health, and the impact on families and society.

What is the molecular characterisation of hereditary hearing loss? How can we use this to target treatment to individual patients?

What is the prevalence and socioeconomic impact of hearing loss in the elderly in the UK?

What is the prevalence and socioeconomic impact of balance problems in the elderly in the UK?

What is the impact of hearing loss on older people with regard to mental and cognitive health, quality of life and the ability to participate in society?

What is the burden on family, carers and society of dizziness and falls in the older adult?

What are the mechanisms behind the link between hearing loss in the elderly and dementia in the elderly?

How can we identify and target patients at risk of age-related hearing loss?

Is loss of sensory function (hearing, smell) a predictor of age-related neurodegenerative diseases or Alzheimer's?

What is the burden and impact of balance disorders at an individual and population level? This includes the socioeconomic impact on ability to participate in work to the individual and society.

What are the causes and course of balance problems, in particular Meniere's Disease?

What is the cause and course of childhood ENT conditions? The conditions stated are
- glue ear,
- recurrent infections: middle ear infections, tonsillitis, rhinosinusitis,
- sensorineural hearing loss,
- sleep disordered breathing and
- nasal obstruction and allergies.

What is the impact of these childhood ENT conditions on the development and educational achievement of children, and on their families and society? What is the impact on otherwise healthy children and in those with complex needs?

What is the prevalence of paediatric balance problems and their impact on learning and locomotor ability? Can the impact be lessened if they are identified early?

What is the prevalence of paediatric tinnitus in different age groups?

What is the prevalence of unmanaged hearing loss in children immigrating to the UK and how should this be managed?

What are the causes and burden of disease of globus pharyngeus?

What are the causes obstructive sleep apnoea and its impact on the health, safety and employment of patients??

What is the impact of facial disfigurement, especially facial nerve palsy, on social isolation and withdrawal?

Define the different variations of disease that make up rhinosinusitis ("endotypes and phenotypes") and develop different ways of classifying them based on signs, symptoms and biochemical markers in order to target them with effective therapy and management.

What are the causes and course of chronic ENT infections in adults with and without treatment, and can we determine what predicts their outcomes? Consider risk factors and improving understanding of inflammatory pathways.

What is the impact on quality of life, relationships, mental health and wellbeing and daily functioning of the loss of sense and smell and taste?

What are the roles of chronic nasal conditions and their management on asthma and lower airways disease?

2. Prevention of ill health and maintenance of good health

How do we prevent hearing loss and tinnitus from developing? What is the risk of personal listening devices and environmental noise?

How can we prevent hearing loss developing from ototoxic drugs?

Developing an adult hearing screening programme: consider practical and cost-effective methods for this, its impact on older people maintaining good health and function, and the feasibility of combining it with a dementia screening programme.

Can self-management strategies be developed and used to improve OSA and snoring?

What lifestyle or environmental strategies can be used to prevent the development of chronic ENT infections and inflammatory conditions, such as the role of diet, supplements and pollution?

What is the clinical and cost-effectiveness of hearing aids for mild-moderate hearing loss?

What are effective rehabilitation strategies for single-sided deafness?

What is the optimal management strategy for the patient with a vestibular schwannoma/acoustic neuroma?

What are the best management strategies for tinnitus?

What is the evidence for auditory implant devices? For example, what are the benefits of cochlear implants compared to middle ear implants in patients with severe (not profound) hearing loss?

Which hearing aid strategies work best? Consider early intervention, user-friendly devices to improve adherence, and the role of emerging communication technology in hearing (such as digital communication devices, technology for self-fitting of hearing aids or brain-computer interfaces for controlling cochlear implants and hearing aids).

What alternative assessments should be used to test hearing beyond conventional Pure Tone Audiograms (PTAs)?

Can we develop effective medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?

What are effective interventions for hearing loss in an ageing population?

How can we restore hearing in older people so that hearing aids are not needed?

What are effective interventions for age-related balance problems in an ageing population?

What is the most effective way of assessing and managing the ENT manifestations, such as loss of smell, voice, swallow, of age-related neurodegenerative diseases?

Develop clear diagnostic criteria and more sensitive tests, including cognitive processing tests, for the diagnosis of vertigo and balance disorders, including Meniere's Disease and vestibular migraine.

What is the role of diet and pharmaceutical drugs in restoring quality of life and independence for patients with dizziness and imbalance?

What is the role and impact of vestibular rehabilitation, including vestibular exercises, on the quality of life, mental health and wellbeing and independence of patients with dizziness and imbalance? Which patients benefit and what are the indications for referral?

What is the best way to evaluate and manage children with tinnitus?

What is the best way to evaluate and manage children with balance disorders?

Develop clearly defined and agreed outcome measures to develop core outcome sets for childhood ENT conditions.

What are the effects of the use (and overuse) of antibiotics in children with recurrent ENT infections? What is the short and long term impact of reducing surgical rates in these children? What is the best management of children with recurrent middle ear infections and is there a role for diagnostic aspiration of middle ear fluid?

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How can we improve the clinical care and safety of patients with tracheostomies both in hospital and in the community? Consider decannulation protocols and tracheostomy tube design.

What is the role for improved diagnostics for endoscopy (e.g. confocal imaging, contact endoscopy, narrow band) in head and neck cancer, and how can emerging diagnostic technology be used in the screening and management of head and neck cancer?

What innovative therapies for facial nerve palsies are clinically effective?

What is the optimal management for adults with OSA? Consider diagnosis, treatment, and its impact on general health.

What is the optimal management (investigation and treatment) protocol for globus pharyngeus?

What is the optimal management for adults with voice problems, including the role of voice rest, diet and pharmaceutical drugs?

Develop clearly defined and agreed outcome measures to develop core outcome sets for benign head, neck and airway conditions.

What are the diagnostic criteria for different subtypes of rhinosinusitis and can these be used to target specific therapies and identify "treatable causes?"

What are the most clinical and cost effective treatments for chronic rhinosinusitis? What is the optimal medical treatment (antibiotics, topical therapies, oral steroids in primary care, immunotherapy), surgical treatments (timing and extent and techniques of surgery, including balloon sinuplasty), or combination of the two for different subtypes?

What is the role for septoplasty or septorhinoplasty in patients with blocked noses?

Develop clearly defined and agreed outcome measures in primary and secondary care to develop core outcome sets for chronic ENT infections, inflammatory conditions and nasal surgery.

4. Designing and Delivering Healthcare

What is the appropriate balance between primary/community and secondary care for different ENT/Hearing & Balance conditions?

What can be safely and effectively managed in primary care, or by patients themselves, and would this be acceptable to patients?

What is already known about designing and delivering healthcare from other fields, and can this be adapted for ENT, Hearing and Balance care?

What is the optimum "system" to identify and treat those who would most benefit from hearing aids or aids to audition?

What is the optimum patient pathway for tinnitus?

What is the optimum design for "balance pathways" (including vestibular rehabilitation) in older adults?

How can GPs and hospital specialists best manage ENT conditions in older people with multiple medical problems?

How can we improve access to hearing services for older adults who are housebound or in residential care, and for those with dementia?

What is the optimum pathway or population-based service for the patient with balance problems? Is there a role for "dizzy clinics," Which specialties should be involved in them and where should they be based?

What is the impact of long waiting lists to access services on quality of life and mental health for patients with long-term vestibular and balance problems?

What age-appropriate pathways can be developed for different types of hearing impairment that reflect the needs and development of children with and without complex needs?

Develop a patient pathway for people with voice problems.

Develop a tracheostomy care pathway across hospital and community settings.

What are the barriers to accessing services for patients with smell and taste disorders? What is the impact of these barriers and delays in referral on these patients?

What is the best pathway for patients with nasal allergies, and do we need to improve access to immunotherapy services? What is the role of multidisciplinary allergy clinics?

How do we implement guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden in adults and children?

What are the variations in adherence management guidelines for rhinosinusitis (EPOS guidelines) in primary and secondary care, and how do we improve their implementation?

What is the variation in management of chronic ear discharge in primary and secondary care, and how do we address this?

6.   Creating effective communication

How can healthcare professionals improve their understanding and communication with patients with tinnitus?

What is the impact of individual and group support networks on patient outcomes for tinnitus and hearing loss? Outcomes include psychosocial and condition-specific outcomes. Developing and assessing shared decision-making tools for parents and clinicians.

Assess communication in multidisciplinary care teams and its impact on quality of care.

How can healthcare professionals improve their understanding of and communication with patients with smell and taste disorders?

What is the impact of anosmia support groups on the health and wellbeing of patients?

How do we standardise consent for nasal procedures such as polypectomy?

5.   Promoting knowledge and learning

Would raising public awareness of the "hidden" conditions of hearing impairment and tinnitus improve the quality of life and wellbeing of people with these conditions?

What is the impact of raising public and professional awareness of hearing and balance problems as more than just a condition associated with age and infirmity? Consider the impact on health behaviours of patients, clinical behaviours of professionals and how they relate to long-term patient outcomes.

What is the impact on patient care, safety and referral rate of training GPs in the management of vertigo, and what is the best way of providing this training?

What is the impact on patient safety of raising public awareness of balance conditions?

How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professional in different settings?

What is the impact of raising public and professional awareness of OSA? Consider the impact on health behaviours of patients, clinical behaviours of professionals and how they relate to long-term patient outcomes.

What is the role of pharmacists in promoting understanding and giving advice about using medications for ENT infections and inflammatory conditions, especially topical nasal sprays?

How can we raise public and professional awareness of the "hidden" problem of anosmia? Consider the impact on health behaviours of patients, clinical behaviours of professionals and how they relate to long-term patient outcomes.

Glossary of Terms

Acoustic neuroma	A benign brain tumour of one of the nerves connecting the inner ear to the brain (see also vestibular schwannoma)
Acute rhinosinusitis (ARS)	Acute infection or inflammation of the nose and sinuses
Adenotonsillectomy	Surgical removal of the adenoids and tonsils
Aetiology	The study of causation of disease
Allergic rhinitis	Inflammation of the lining of the nose caused by allergies
Anosmia	Loss of sense of smell
AOM (acute otitis media)	Middle ear infection
Audiogram	The graph that shows the results of a hearing test
Audiology	A branch of science that studies hearing, balance and related disorders
Auditory implant devices	Surgically implanted devices to restore or amplify hearing
Chronic rhinosinusitis (CRS)	Chronic inflammation of the nose and sinuses sometimes leading to the development of polyps
Cochlear implant (CI)	A surgically implanted device that restores partial hearing to people with severe to profound hearing loss
Decannulation	Removal of a tube, such as a tracheostomy tube
Dysphonia	Disorder of the voice
Endoscope/endoscopy	A tube with a light and a camera to look inside the body to examine or diagnose conditions
Endotypes	A subtype of a condition, caused by different mechanisms
ENT	Ear, Nose and Throat surgery
Epley manoeuvre	A positioning manoeuvre to treat BPPV
EPOS	European Position Paper on Rhinosinusitis and Nasal Polyps
Globus pharyngeus	Subjective feeling of a lump or foreign body in the throat
Glue ear (OME)	A common childhood condition in which the middle ear becomes filled with fluid (otitis media with effusion)
Grommets	Small tubes inserted into the eardrum to drain middle ear fluid
Incidence	The number of new cases of a disease or condition per population at risk in a given time period
Laryngectomy	An operation to remove the larynx (voice box)
Mastoiditis	Acute or chronic infection/inflammation of the mastoid bone behind the ear

Meniere's disease	A disorder causing vertigo, tinnitus, hearing loss and a feeling of fullness in the ear
Nasal Polypectomy	Surgical procedure to remove polyps from the nose and sinuses
Obstructive sleep apnoea (OSA)	A sleep disorder where breathing stops at least 10 seconds every hour during sleep characterized by recurrent episodes of upper airway collapse
Otolaryngology	The area of medicine dealing with ear, nose and throat conditions and management of cancers of the head and neck
Ototoxic/Ototoxicity	The property of being toxic to the ear
Pathogenesis	The biological mechanisms that lead to a disease or condition
Phenotype	The observable characteristics of a condition, used to classify diseases
PPI	Patient and Public involvement
Prevalence	The proportion of cases of a disease or condition in the population at a given time
Pure tone Audiogram (PTA)	A pure-tone air conduction hearing test determines the faintest tones a person can hear at selected pitches (frequencies), from low to high. During this test, earphones are worn so that information can be obtained for each ear. The results are recorded in an audiogram.
Rhinology	The study of conditions of the nose and sinuses
Rhinosinusitis	Inflammation or infection of the lining of the nose and sinuses
Sensorineural hearing loss	Hearing loss due to damage to the inner ear or nerve pathways from the inner ear to the brain
Septoplasty	Surgical procedure to straighten the septum of the nose
Septorhinoplasty	Surgical procedure to straighten the septum and bones of the nose
Tinnitus	The perception of sound in the ear when no external sound is present, also known as "ringing in the ear"
Vestibular	Relating to the sensory system that detects balance and spatial orientation
Vestibular schwannoma	A benign brain tumour of one of the nerves connecting the inner ear to the brain (see acoustic neuroma)

Appendix 12: Consensus Conference small group facilitator briefing guide

GENERATE

Towards a National Research Agenda for ENT, Hearing and Balance Care

Consensus Conference Briefing Document for Small Group Facilitating

13.30 – 17.30, 25th September 2015

The Farr Institute, London

Consensus Conference: Preparation and Format for the Day

GENERATE has been initiated to develop a research agenda for ENT, Hearing and Balance care in the UK. It is a groundbreaking project that has been shaped by the views of patients, carers, healthcare professionals, researchers and other people that develop, use and benefit from evidence for clinical care.

We have gathered a list of research ideas that these people think should be considered for the final research agenda. The task for the Consensus Conference participants will be to decide on the ideas that should form the basis for the research agenda for ENT, Hearing and Balance care. We will not be trying to draft formal research questions or the full agenda on the day.

In order to decide the research agenda we will be asking the stakeholders:

4. **To discuss what they think are the most important research ideas to be included in the agenda**
5. **Why they think these are important**
6. **To come to an agreement as a group at the end of the Conference as to what should be included in the final research agenda.**

These discussions will take place both in a large group and in smaller groups to discuss the specific topic areas in more depth (see Figure 1 below).

Conference starts: session with all participants facilitated by Phil Hadridge.

Small group sessions: facilitated sessions to choose topics in each group. Refreshment break provided.

Conference closes: Session with all participants facilitated by Phil Hadridge.
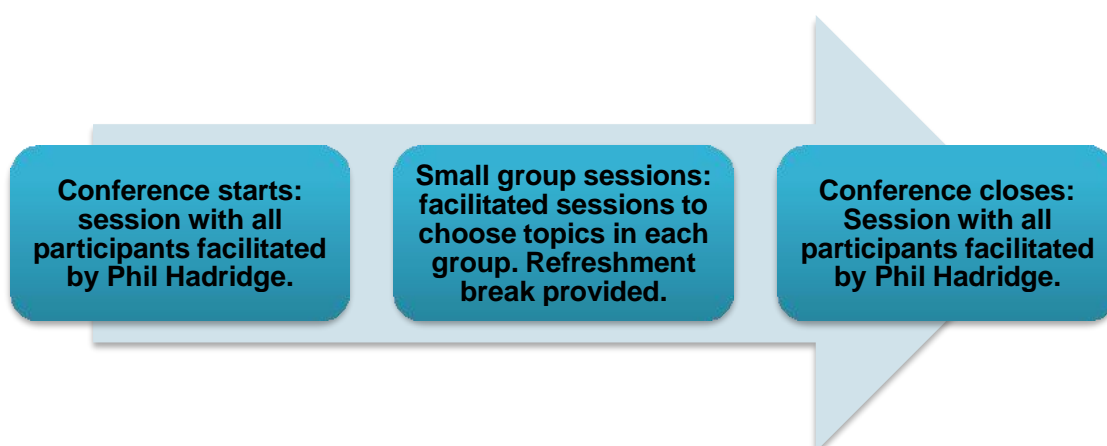
We have asked the participants to look at the questions in the topic they have been allocated to in more detail before the conference and consider which questions should form part of the research agenda and why they should be included. They will select two top choices and two more as your second choices and will be asked to feedback on this in the small group sessions.

<u>**The role of the facilitator**</u>

The aim of the small group work is to come to consensus about which questions to present to the final plenary session as their choices to be part of the agreed research agenda. The small group sessions will last 90 minutes.

These groups are a mix of clinicians, professionals and patient/lay representatives. Some will be more familiar and comfortable with research than others. Please be aware of this in the group dynamics to ensure that traditional "experts" do not dominate the discussion. Everyone's experience in these groups is of equal value. It is our responsibility as facilitators to help encourage all members within the small group to discuss their decision-making in choosing their top choices based on:

1. **What they think are the most important questions to be included in the agenda.**
2. **Why the participants think these are important.**

As most of the questions are fairly broad it may be reasonable to assume that the participants may see the broad themes as a priority setting. However, ideally we are looking to identify specific questions from the themes.

The participants will be asked to decide their top two questions (and reserve two questions) for their group domain in advance – this will help the sharing in during the small group discussion.

<u>**Outline of small group work:**</u>
Start session with brief ground rules: respect for beliefs, disagreement is OK, only talk one at a time, be aware of any communication issues, be prepared to listen and discuss. This is not about "right and wrong" it is about negotiation. Acknowledge these are hard choices.

Ask for brief intro from each participant (name and role) and for their top two + reasons. Make a note of these on the visual aids provided. This will be followed by facilitated group discussion to agree the top two questions and top two reserve questions. When these have been agreed, please use the voting cards provided to check degree of consensus (see below). Please record this result on the voting record sheet provided.

Visual aids:

A3 sheet with the whole list of questions in your topic: questions that the group agree are important can be highlighted on these, and questions that the group agree are less important can be crossed through.

Large poster to display the key questions in your topic: the questions that the groups feel are the most important can be written on here with reasons for their importance. At the end of the session the top two questions can be highlighted.

Voting cards: these can be used if consensus cannot be reached by discussion, and to test degree of consensus with the final questions chosen. You may be providing with coloured cards e.g. Green cards for top questions and orange cards for reserved.

The A3 sheets and large poster are designed to be written on, so please encourage your group to display their work visually as the discussion progresses. At the end of the small group session there will be a coffee break and the posters will be displayed to the whole group as a form of peer review before the final discussion.

Voting record sheets:

You will be provided with a voting record sheet to record the results of the final vote to check consensus in your group. Please fill this out and return it to Ilia Papachristou.

What degree of consensus do we need?

There is bound to be disagreement. Therefore, if the groups cannot agree through negotiation we will look at voting strategies for a majority. Again, using visual aids, such as voting cards.

We will audio tape all small group discussions so you are not expected to take notes. Although we will ask you for some feedback on how you feel the session went and any outstanding group dynamics that you felt influenced the discussion.

The full meeting agenda will consist of:

<u>Four hour conference</u>

30 minutes warm up introduction with a plenary session
90 minutes small group focused discussion. This is the session you will be facilitating.
30 minutes comfort break (teas and coffees). Posters will be displayed here for all groups to see where each other has got up to.
90 minutes for final plenary group session.

Top tips from Professional Facilitator Phil Hadridge

Remember the four facilitation roles
Please keep in mind that there are four key areas that will help a group get further:

1. Keep the group to **task**

2. Keep the session to **time**

3. **Include** all participants in the discussion (including through voting, writing individual ideas on pieces of paper)

4. Ensure the ideas are **captured** and fed in (or fed back)

You might want to 'delegate' the timekeeping and capturing roles.

Be clear on the question
A short, concise question is a powerful way of framing the purpose/task of a session and the outcomes you are looking to achieve. Consider these four dimensions in forming a question:

a) What is the best way to start: "In light of…"; "Given,"; "What…"
b) Which framing word is most useful: what, how, why, where, when who?
c) What 'power' words open up the mind and get the creative juices going: critical, simplest, significant, challenging, relevant… or any other adjectives can be used.
d) What sense of action or follow-through do you want to infer with the question?

First things are fateful
My principle is that everyone uses their own voice in the first few minutes of any session (maybe in a paired conversation or going round the participants or…). This helps to settle the group – which is especially important if there are some speakers to listen to before any group work.

What risks do you fear?
Common worries can include

- Silence (or too many trying to talk!)
- Not enough time (an over ambitious agenda)
- A flat vibe
- Competition between participants
- A tricky and hard to handle issue lurking 'in the corner'
- Honesty discussion diminished as the group is daunted by a powerful person present
- Group think in discussion or decision making
- Interaction and voting imply democratic decision making

Appendix 13: The research priorities with examples from the survey data

Priority	Example of source data from survey
Define the different variations of disease that make up rhinosinusitis (endotypes and phenotypes) and develop ways of classifying them based on signs, symptoms and biochemical markers.	"1.What causes sense of smell and taste to deteriorate? 2.How can the symptoms of loss of smell/taste be targetted to sufferers?" (respondent 189, patient) "Treatment of chronic rhinosinusitis … CRS is common and major source of poor QOL and performance. Treatment options including steroids, macrolides and surgery with little evidence either way. Urgent need for proper phenotyping of these patients" (respondent 153, clinical academic)
What are the most clinical and cost effective treatments for chronic rhinosinusitis?	"I have lost 95% of my sense of smell and taste now for 2 and a half years following a severe bout of Bronchitis. After receiving treatment from my Doctor and visits to ENT hospital in Inverness they tell me know that there is no more they are able to do. The treatment prescribed mainly consisted of steroid tablets, drops and nasal sprays. I have had 2 CT scans and the examination shows no signs of polyps. They feel that an operation on my sinus is not required at present. In addition to this I have undertaken a course of acupuncture and herbal medicine which has I incurred almost £600 of expense. I feel strongly that further research is urgently required in the smell and taste fields as a knowledge of treatment is very much lacking at present." (respondent 122, patient) "Should all patients who are undergoing FESS for rhinosinusitis with polyps have graduated surgery based on extent of their disease or should they proceed to complete FESS with a Modified Lothrop procedure?" (respondent 159, ENT surgeon)
How do we improve the implementation of guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden and its impact on antibiotic resistance in adults and children?	"…Recurrent AOM and antibiotic sparing approaches… Treatments are problematic especially antibiotics (resistance) and there is still more to be done finding evidence based treatments…" (respondent 101, GP). "How do we effectively minimise the antibiotic burden in children with upper respiratory disorders? How can ENT surgeons best influence management of conditions treated predominantly by other specialties (especially general practice) What are the best strategies to reduce/avoid emergency admissions in ENT?...Issues affecting a large proportion of society (community patients, children and emergency attendees), with wide-reaching effects (antimicrobial resistance, practice variation, costs to NHS). These are also areas linking with the NHS Outcomes Framework and Department of Health priorities." (respondent 502, clinical academic)
What outcome measures and instruments should we use in primary and secondary care for chronic ENT infections and inflammatory conditions?	"As a GP my perspective is better management of common ENT disorders. They are so frequent!... Better integrated management e.g. of OME. Which outcomes are most important (agreement) and how can these be used in developing a rational/ good chronic disease model of care (cf asthma care)" (respondent 101, GP)
What is the optimal management for adults with OSA?	"Snoring and sleep medicine…There is also limited evidence base for snoring surgery." (respondent 321, ENT surgeon). "Snoring research. Affects a great number of people across the uk in their ability to conduct a normal life through disturbed sleep. Inability to maintain a normal relationship with partners due to Seperate bedrooms." (respondent 518, patient)

What outcome measures and instruments should we use for research into head, neck and airway disease, both benign and malignant?	"More help for throat cancer patients, particularly Laryngectomees. There must be ways of re-installing sense of smell, and taste, after throat operations, with new drugs/treatments..? Also, I know of lot of great work is being done in Stem Cell/Transplant research/tests for pre-op potential Laryngectomee patients. But what about 'established' Laryngectomees..? I think research should also be targeted to this group, with maybe new surgery to install an artificial windpipe to the mouth area, which would help allieviate some of the mucus problems that Laryngectomees face today, and do away with neck breathing..." (respondent 118, patient) "Accurate survival, functional and quality of life outcome measures for head and neck cancers to advise patients, clinicians and decision making." (respondent 46, ENT surgeon)
What is the role for improved diagnostics in head, neck and airway conditions?	"Globus pharyngeus represents a massive burden on the NHS. Secondary care only sees the tip of the iceberg. I work in community ENT where there are a significant number of patients with this condition. Most have spent years visiting their GPs and undergoing tests (chest x rays, barium swallows, gastroscopies, ultrasound scans of the neck, etc) as well as receiving referrals to chest physicians for chronic coughs then undergoing more potential tests such as bronchoscopies and CT scans of the chest. The link betweek bruxism, laryngopharyngeal reflux, stress and globus pharyngeus needs further research. The causes of the condition need to be determined with confidence. Treatment guidelines need to be issued to reduce unnecessary investigations and to provide patients with a useful treatment plan. To continue to claim, as a speciality, that the aetiology is 'unknown', and to inform patients that 'nothing is seriously wrong', does not address the patients concerns or allow for any sensible treatment plans. Massive burden of disease/illness in patients that receive no useful diagnosis or treatment from primary or secondary care. Massive expenses to the NHS in repeat appointments and recurrent investigations, without a conclusion every being reached." (respondent 124, ENT surgeon)
What is the optimal management for adults with voice problems?	"What are the best methods to assess and treat voice disorders? Because there are different methods used by speech therapists and voice clinics across the UK and not enough evidence on the efficacy of therapy." (respondent 107, speech and language therapist) "Problems with hearing, balance and voice may not be life threatening but are most certainly quality of life limiting. I would vote for more research into what social and technology support can be designed to help overcome these confidence sapping issues before they cause unnecessary isolation and restrict activities of daily life. Social interaction and communication with others is vital for humans to function with dignity and independence." (respondent 418, patient)
What is the optimum pathway for people with balance problems? What can be dealt with in primary care and the community, and what are the indications for referral? How should specialist clinics be set up?	"Definite primary care pathway for treatment of the dizzy patient… To reduce inappropriate referrals to ENT for dizzy patients." (respondent 267, nurse) "Balance big problem for general practice patients. Doesn't seem to be a clear pathway for what to do with these patients or what works." (respondent 374, GP)
How can patients be empowered to self-manage their balance problems and how does this impact on their outcomes?	"To design a survey to diagnose the nature of a balance problem on-line because ENT doctors are very busy and this might speed things up. To design an individual exercise programme on-line because the patient needs ongoing input and encouragement and the person giving the exercises is

	also very busy To find out the many causes of vertigo and the factors which make things worse all the time because it takes a long time to discover everything which might affect the balance." (survey respondent 656, patient)
Develop clear diagnostic criteria for different balance disorders so that patients can be informed about prognosis and best treatment.	"Vestibular/Vertigo-related matters are of a personal concern. Sudden onset apparent BPPV followed by a misdiagnosis of Meniere's Disease with evential diagnosis of Vestibular Migraine…Quick, accurate diagnosis by NHS specialists - not private consultations The vertigo first displayed itself very suddenly three years ago, following an investigated (privately) but undiagnosed pain in the side of the head which got worse on movement not on touch. Then a sharp gust of wind into the ear, followed within days by severe dizzy spells on movement of the head. Again, privately, I was treated for BPPV and this seemed to work. But then a couple of months later, I was very dizzy and very sick. I was due an appt but this was some weeks ahead and it was not possible to bring it forward on the NHS. I had to wait. In the winter of 2011 I had such severe vertigo that I was knocked off my feet and it took two weeks to learn to walk again once I'd stopped being sick. Investigations were undertaken by local NHS ENT who initially suggested Meniere's Disease and this caused a stir at work where I was no longer allowed to park in the company ar park... and other discriminatory behaviour. After hot and cold water in the ears and other gross tests I was told that I had vestibular migraine and discharged... No follow up. No progress check. No ermegency number should I get a sudden attack... I live in dread of it happening all over again." (respondent 68, patient).
What are the best management strategies for tinnitus?	"What is the value of Tinnitus Clinics in terms of improving the quality of life in these patients? Tinnitus is commonly seen in Primary Care and yet management strategies to help these patients are often limited." (survey respondent 665, commissioner)
Which hearing aid strategies work best for adults with hearing loss?	"All ENT depts should automatically consider other options such as BAHAs; digital hearing aids; sign language classes lipreading classes; deaf awareness for all ENT front line & backroom staff instead on constantly pushing cochlear implants as the only solution to deafness in children and adults. Cochlear implants seem to get a lot of research and development funding but this shouldn't be the case as the Deaf community is very diverse and all options should be researched equally." (respondent 62, not stated)
What is the burden and impact of hearing loss on an individual, family and population level?	"Aging population is increasing and so there are some concerns that undiagnosed hearing loss will increases cognitive decline and dementia- can early diagnoses reduce this risk?" (respondent 51, audiologist)
What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?	"How do we restore hair cell and neural function (stem cells? genetic therapies? pharmaceuticals?)… although hearing aids and cochlear implants are important management options, they are not cures, and do not restore hearing ability fully. We need to be working towards a cure for hearing loss, repairing the damaged tissues." (respondent 76, academic)
What are effective interventions for hearing loss in	"Progressive hearing loss with age. It affects so many people and makes it difficult for older people

older people?	to participate in normal human activities, which is a sad way to spend the last years of life. It leads to many miscommunications and arguments." (respondent 65, academic)
Is it beneficial to introduce an adult hearing screening programme and how do we best develop it?	"Please support a general hearing test at 60-65 years of age. I have...and I have, hundreds of elderly people asking me how I cope with hearing aids. But most of them left it too late, and could not cope with the introduction. Equally.....they were usually very short changed by audiology...with the test, and then a hearing aid shoved in there lug...and that was it. No offer of adjustments whatsoever. Most elderly folks found the additional noise....weel...just to noisy....and the hearing aid is now not used. Sad! This is a very poor service." (respondent 59, nurse)
What is the optimum design for "balance pathways" in older people with balance problems? How and where can older people with balance problems and with multiple medical conditions and decreased mobility best be managed? How can self-management strategies be built into these pathways and what is their impact on patient outcome?	"I am a recent patient and am here for a minor ailment. However, my general concerns about ENT, hearing and balance is the deterioration of sight and hearing as one grows older. As someone who has had a weaker eye since childhood I was only told recently that my sight would deteriorate dramatically with time unless I wore corrective glasses and updated the prescription regularly. As a healthy male approaching his 30s I would like to have more information about the possible changes in sight, hearing and balance and so would like further clinical research to focus on this. This seems important to me because hearing, sight, balance are hugely important to me, as they are to everyone. I would like to look after my senses, sight and hearing as best I can as I grow old so would like more information to be readily available." (respondent 9, patient)
What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?	"Diagnostic tool for APD. Effective management of APD. Sensitive outcome measures of non-technical interventions. Improved treatment. Ability to seperate APD individuals from those with learning problems etc Ability to provide treatment/advice with a known efficacy." (respondent 628, hearing therapist)
How do we best diagnose and manage obstructive sleep apnoea (OSA) in children?	"Management of glue ear in children. Management of allergic rhinitis Prevention of recurrent tonsillitis Outcomes of adenotonsillectomy for Obstructive sleep apnoea in children. These are all very common conditions of childhood which have a significant impact on the child a short and long term as well as their educational attainment." (respondent 58, ENT surgeon)
How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professionals in different settings, and what is the impact of this on patient outcome?	"There doesn't seem to be much weight given to the life stage of hearing loss A hearing loss from birth has ALOT more affect than a mild age related loss from 50 on. Both in terms of impairment duration and 'importance'. Eg Childhood / Education and employment are affected if hearing impaired from birth. Hearing loss experienced at 50 on is post most of the above events. Yet - ENT handles HI at younger ages the same as OAPs." (respondent 68, patient)

Table 35 The research priorities with examples from source survey data

Appendix 14: GENERATE final agenda

(Bohm *et al* 2015)

The Research Agenda for ENT, Hearing and Balance Care
A UK Partnership of Patients, Professionals and the Public

GENERATE
Patients, Professionals, and the
Public in Partnership

Acknowledgements

Contents

365

Foreword

We are delighted to present the national research agenda for ENT, Hearing and Balance care in the UK. This document summarises the results of three years of planning and working with patients, the public and our professional colleagues from across the UK to decide which questions around the care of people with ENT, Hearing and Balance problems are most in need of research. We would like to thank all the participants who have contributed their ideas and shared their time, many of whom travelled long distances to take part in the discussions. We hope that you can hear your own voices clearly in this research agenda. This agenda is only the start of the journey to develop the evidence base for ENT, Hearing and Balance care. The partnerships built through this initiative will be carried forward in order to deliver this agenda and develop high value research and care for the benefit of our patients and their families.

Professor Anne Schilder
Professor Valerie Lund CBE
Ms Natalie Bohm

Introduction

We strive to deliver the best care for patients with ENT, Hearing and Balance conditions. To do this, it is key that high value research is developed and conducted to inform us about what treatments work best and how we can most effectively deliver care. Recognising that competition for research resources is strong, and that ENT, Hearing and Balance professionals need to build capacity and expertise to deliver the research needed, ENT-UK took the initiative to develop a strategic research agenda. With the support of the British Society of Audiology and the British Academy of Audiology, ENT-UK commissioned the evidENT team at University College London (UCL) to conduct a research priority setting exercise aimed at identifying research priorities in ENT, Hearing and Balance. This initiative was named GENERATE, and the research agenda presented in this booklet is the result of this collaborative work.

Methodology

In January 2014, an independent Steering Committee was established to oversee GENERATE. This Committee included representatives of the groups that develop, use or benefit from research in ENT, Hearing and Balance. evidENT, a multidisciplinary clinical research team based at the Ear Institute at UCL and supported by the National Institute of Health Research, worked with academics from UCL's Department of Applied Health Research and Improvement Science London to design the research priority setting exercise. The approach used was based on a systematic review of the literature on research priority setting in healthcare. Research ideas were gathered through an online survey posted through patient and professional groups. Respondents to the survey were asked to indicate, based on their experience, the most important topics, questions or issues in ENT, Hearing and Balance care that should be answered by research, and why they felt these were important. In addition, five focus groups involving patients, healthcare professionals from various disciplines and settings, service managers and commissioners, researchers and representatives from industry, met to discuss these questions in more depth. These discussions were audio recorded and transcribed to ensure that all information was captured. Many of the issues and research ideas raised in the survey and focus groups were not traditional research questions, yet these were the issues that our respondents felt were important. Therefore a
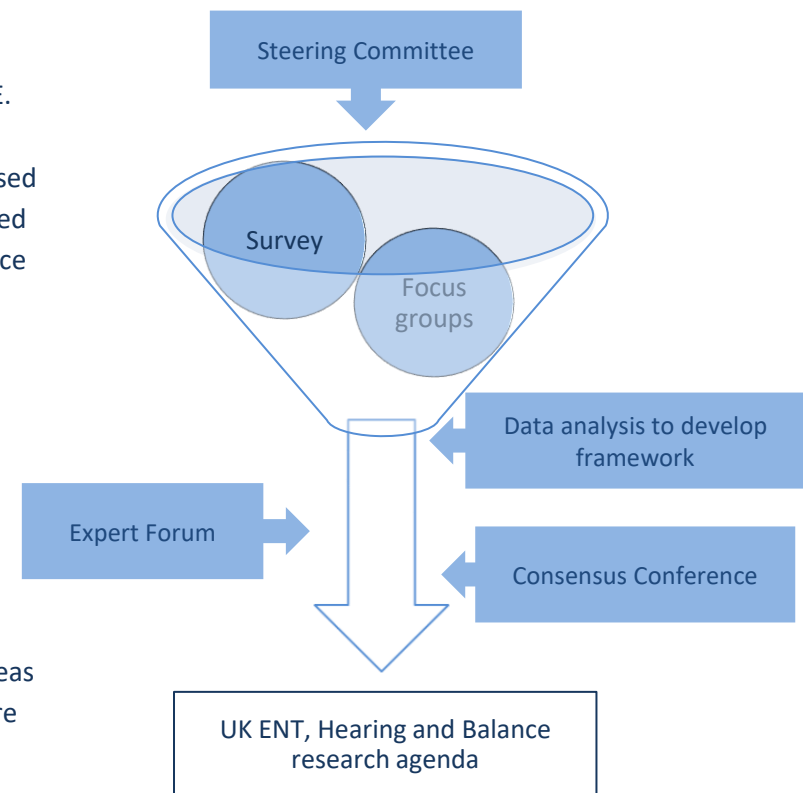
framework analysis[2] was used, allowing all those ideas to be incorporated into the agenda. Using this method the academic team analysed the data and identified a number of research themes and topics into which all the ideas could be categorised (these form the framework and are described in detail below). Initial analysis produced a long list of research ideas across this framework. An Expert Forum of 21 participants, chosen based on the framework themes and topics, and including patient participants, was held in June 2015 to put the research ideas into the context of current evidence and developments. This led to a shorter list of research ideas that were prioritised in a Consensus Conference held in September 2015. This Conference had 48 participants including patients, members of the public, healthcare professionals from different specialties and disciplines, research funders and policy makers. They discussed and prioritised the research ideas in facilitated small groups. This was followed by a plenary session where these priorities were reviewed by all participants and final consensus was reached.

Results

There were 685 responses to the survey. Other than the six key healthcare professional groups (audiologists, audiovestibular physicians, ENT surgeons, GPs, nurses and speech and language therapists), there were respondents from 21 professional groups including paediatricians, geriatricians, physiotherapists, clinical psychologists, pharmacists and teachers of the deaf. Non-healthcare professional respondents identified themselves as patients, patient carers, parents, members of the public, voluntary workers and charity organisations. The five focus groups involved 43 people from various backgrounds (see above).

The survey and focus groups raised over 1500 research ideas. After removing duplicated suggestions and further analysis 670 research ideas were identified; these were reviewed in the Expert Forum. The experts advised removing questions that had already been answered or were the subject of on-going research within ENT, Hearing and Balance, or could be answered by using knowledge from other fields. They also advised combining some of the topics into broader topic areas as there was some overlap. As a result, 99 research ideas across six different topic areas were presented to participants in the Consensus Conference. Their discussion resulted in 21 priorities for the research agenda for ENT, Hearing and Balance care.

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[2] Ritchie J, Spencer L and O'Connor W. (2003). Carrying out qualitative analysis. In Ritchie J. and Lewis J., eds. *Qualitative Research Practice* London: Sage Publications Ltd. Ch. 9.

The GENERATE Framework

An overview of the framework for the research agenda in ENT, Hearing and Balance care is presented on page 10. A list of all initial research ideas structured across the framework will be available in the full online report. From the survey responses and focus group discussions, six broad research themes were identified and included in the framework. The definitions of the research themes are listed below. A seventh theme 'Creating a research culture in ENT, Hearing and Balance' contains cross-cutting ideas about what the final agenda should incorporate and strategies for implementing it, rather than research ideas. It is outlined in detail on page 20.

Analysis of the survey responses and focus group discussions revealed that they crossed six topics, based on ENT, Hearing and Balance problems and the different age groups affected by them. The research priorities in this booklet are presented under these research topics for clarity. The full online report will include an overview of the research priorities across the research themes. An explanation of the research topics is on page 9.

Research Themes

Understanding health, illness and disease: What causes ENT, Hearing and Balance problems, and what impact do they have on people, their families and society?

Prevention of ill health, maintenance of good health: How can we prevent ENT, Hearing and Balance problems and keep people healthy?

Clinical and cost-effectiveness: How effective are current and new ways of diagnosing and treating ENT, Hearing and Balance conditions?

Designing and delivering health care: How do we improve the quality and value of ENT, Hearing and Balance care, and make it sustainable and patient-centred?

Creating effective communication: How can we improve communication and interactions with patients and between healthcare professionals in order to improve health?

Promoting knowledge and learning: How can we share knowledge and learning with patients and professionals in order to improve health?

Inflammation, Infection and Allergy in Adults: This topic includes research ideas about allergies and infections of the ears, nose and throat in adults. Children also suffer from these infections, these ideas are captured under the topic 'Children and Young People' Participants noted that despite previous research efforts many uncertainties exist. Policy makers often quote ENT operations for these conditions as an example of practice variation.

Head, Neck and Airway Problems: This topic deals with ENT conditions that affect voice, swallowing and breathing. Clinicians recognised that these increasingly account for time taken in clinic, and many are uncertain about the best ways of managing them. Patients felt that they were receiving inconsistent advice and that there seemed to be a lack of knowledge about best management.

Balance Problems: This topic includes uncertainties raised about diagnosis and treatment of balance conditions across all ages. This area was felt to be under-researched and poorly understood, despite its significant impact on quality of life and day-to-day ability to function. As one survey respondent noted when answering why this should be a priority area: "You wouldn't ask if you had it."

Adult Hearing Loss and Tinnitus: Both patients and professionals felt that more research was needed in hearing loss and tinnitus in adults. The personal and societal impact of hearing loss is vast and under-recognised. Children also suffer from hearing loss and tinnitus, and these ideas are captured under the topic 'Children and Young People.'

Children and Young People: ENT, hearing and balance conditions are common in this age group; many concerns were raised about their impact on children and young people, who are "…yet to live their life" (Patient, survey respondent). Special mention was made of children with complex needs who often have ENT problems but have been excluded from many studies so far.

An Ageing Population: Many issues were raised around the health challenges faced by an ageing population. Loss of ENT, hearing and balance function has a significant impact on the health and wellbeing of older people, and on their families. This also incurs high health care costs to society.

Research Topics	Adult hearing loss and tinnitus	Balance problems	An Ageing Population	Children and young people	Inflammation, infection and allergy in adults	Head, neck and airway problems

Understanding health, illness and disease

Prevention of ill health, maintenance of good health

Clinical and cost-effectiveness

Designing and delivering health care

Creating effective communication

Promoting knowledge and learning

Themes

Values and Criteria for Prioritisation

One of the guiding principles of GENERATE is that it is transparent, both in its process and in its decision-making. Part of this transparency involves understanding the participants' reasons for choosing the research priorities that are summarised in this agenda. All the way through the initiative, from the initial survey and focus groups to the Consensus Conference these criteria, or values, have been explored and discussed. These are summarised in the figure below. Some of the values may seem to contradict each other: prioritise pragmatic research projects versus aspirational projects; focus on conditions that affect large numbers of people versus on those that have a major impact on fewer people; find better ways to use the knowledge we already have or develop entirely new ideas? There is no right or wrong answer in this. Importantly, these options were considered and weighed so that consensus could be reached, even if there was not always perfect agreement.
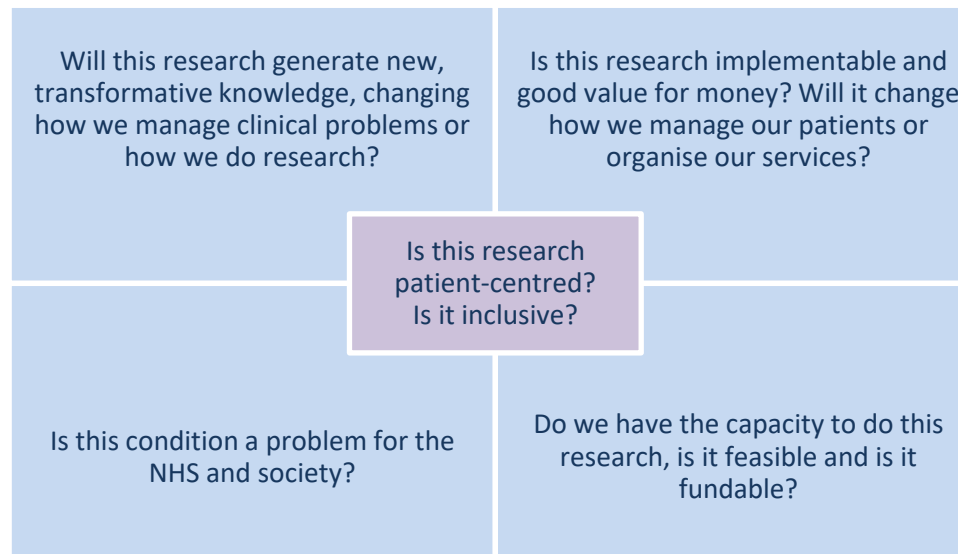
Will this research generate new, transformative knowledge, changing how we manage clinical problems or how we do research?	Is this research implementable and good value for money? Will it change how we manage our patients or organise our services?
Is this condition a problem for the NHS and society?	Do we have the capacity to do this research, is it feasible and is it fundable?

Is this research patient-centred? Is it inclusive?

"...actually are we at the stage of talking about treatments? Shouldn't we focus more on aetiology, causation..." (Researcher, focus group participant)

"...there are so many areas, not only from the technological side but also from beyond the technological side how to develop the services, how to run the services which can be looked into purely from a research point of view" (Healthcare professional, focus group participant)

"I think maybe instead you could do some human research instead of some laboratory research, it would be good..." (Patient, focus group participant)

"These are all very common conditions of childhood which have a significant impact on the child a short and long term as well as their educational attainment" (Healthcare professional, survey respondent)

"...in silence tinnitus is always there to remind you something's wrong, and in noisy environments the deafness is always there to remind you something is wrong, and that's why it's incredibly insidious" (Patient, focus group participant)

"It's not life threatening. It's not visible. However smell and taste are TWO of the senses. This has a huge effect on the quality of life on a day to day, hour by hour, minute to minute basis" (Patient, survey respondent)

Adult Hearing Loss and Tinnitus
- What are the best management strategies for tinnitus?
- Which hearing aid strategies work best for adults with hearing loss?
- What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?
- What is the burden and impact of hearing loss on an individual, family and population level?

Balance Problems
- What is the optimum pathway for people with balance problems?
  - What can be dealt with in primary care and the community, and what are the indications for referral?
  - How should specialist clinics be set up?
- How can patients be empowered to self-manage their balance problems?
- Develop clear diagnostic criteria for different balance disorders so that patients can be informed about prognosis and best treatment.

An Ageing Population
- What are effective interventions for hearing loss in older people?
- Is it beneficial to introduce an adult hearing screening programme and how do we best develop it?
- What is the optimum design for 'balance pathways' in older people with balance problems?
  - How and where can older people with balance problems and with multiple medical conditions and decreased mobility best be managed?
  - How can self-management strategies be built in these pathways and what is their impact on patient outcome?

Children and Young People
- What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?
- How do we best diagnose and manage obstructive sleep apnoea (OSA) in children?
- How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professionals in different settings, and what is the impact of this on patient outcome?

Inflammation, Infection and Allergy in Adults
- Define the different variations of disease that make up rhinosinusitis ("endotypes and phenotypes") and develop ways of classifying them based on signs, symptoms and biochemical markers.
- What are the most clinical and cost effective treatments for chronic rhinosinusitis?
- How do we improve the implementation of guidelines on antibiotic use in ENT infections in order to reduce the antibiotic burden and its impact on antibiotic resistance in adults and children?
- What outcome measures and instruments should we use in primary and secondary care for chronic ENT infections and inflammatory conditions?

Head, Neck and Airway
- What outcome measures and instruments should we use for head, neck and airway conditions for benign and malignant disease?
- What is the optimal management for adults with OSA?
- What is the optimal management for adults with voice problems?
- What is the role for improved diagnostics in head, neck and airway conditions?

Developing a strategic research agenda

This agenda should:
- Ensure that future research in ENT, Hearing and Balance is patient centred and of high value.
- Ensure that research leads to the development of sustainable high value health care in ENT, Hearing and Balance.
- Incorporate novel, innovative therapies with the potential for benefit in the future.

In order to implement this agenda the following strategies need to be considered:
- Develop the capability and capacity for research in ENT, Hearing and Balance by growing the workforce to develop and deliver the projects.
- Develop and nurture research collaborations across both health care and academic disciplines in order to develop and deliver the high value translational research that has the potential to change practice.
- Developing data collection platforms that capture information that is both applicable to clinical care as well as research.
- Clinicians and researchers should work with patients and carers to decide which outcomes to include in future research.

Adult Hearing Loss and Tinnitus

The Priorities

- What are the best management strategies for tinnitus?
- Which hearing aid strategies work best for adults with hearing loss?
- What are the effects and costs of novel medical and biological treatments (such as cell-based therapies) for sensorineural hearing loss?
- What is the burden and impact of hearing loss on an individual, family and population level?

Hearing loss and tinnitus have been research priorities for patients and professionals through the initiative. Both hearing loss and tinnitus are recognised as common conditions that are often not obvious; symptoms may be hidden but can have an impact both on those suffering from it as well as those around them. In particular tinnitus was highlighted by patients as a condition with underestimated impact. Research in this area has been seen to be under-resourced, with a lack of evidence for current management strategies. Research into the effects and costs of existing and novel therapies for tinnitus has the potential to provide benefit to patients and to the NHS by reducing the burden of tinnitus.

As noted in other topics, hearing loss is a growing burden on society and the NHS; it may have a significant impact on people's mental and cognitive health and ability to interact in society. Hearing loss is a condition of major health, societal and financial impact. It affects people's ability to communicate and function in the workplace, and has been linked to dementia.

Although hearing aids continue to improve, their uptake is low. More research into technologies to improve their user-friendliness could provide a rapid return in terms of patient benefit and research investment. Implantable devices to improve hearing, such as cochlear implants and middle ear implants, were also considered under "hearing aids." With rapid advances made in this field, it was noted that research into who would benefit from them, and when, would be timely. Registries of patients receiving such implantable devices would facilitate such research.

Investment in research that was aspirational in terms of being potentially years away from application in clinical practice was also considered alongside research with more immediate benefit to patients. The development of novel, innovative therapies for sensorineural hearing loss was recognised as having the potential to transform care in the future, and therefore was regarded as one of the research priorities.

Better understanding of the burden and impact of hearing loss on an individual, family and at a population level is important to both patients and professionals. Although research has been done in this area it was felt that more is needed to raise the profile

Reasons for these choices

Tinnitus is a common, often hidden condition with underestimated impact; research has been under-resourced and there is a lack of evidence for best management.

Hearing loss is a common, growing problem affecting large numbers of people.

The uptake of hearing aids is low and many people are unhappy with the benefit they offer; research into new devices and technologies would help us better meet population needs and could provide rapid benefits.

Long-term aspirational priorities, such as research into novel hearing treatments should balance shorter-term research gains.

The better the understanding of burden and impact of hearing loss, the more value research and services based on this understanding will have, as it will be informed by patient experience and clinical need.

of the problem with policy-makers, research funders and service commissioners. This knowledge would inform future research and health services, basing it around patient experience and clinical need, as well as providing the incentive for resource allocation.

There has been clear recognition throughout the initiative that access to services is a concern for people with balance problems: access to the right care by the right specialists, access to support and access to knowledge about management options. Balance problems are known to be common and may have a significant impact on the lives of people of all ages, yet survey respondents and focus group participants expressed concerns about poor management with long delays in diagnosis and treatment. This delay causes significant stress to sufferers, with patients moving from clinic to clinic until they get a diagnosis. Service organisation was raised as key to alleviating this. The research priorities reflect this need, calling for research into the optimum pathway for people with balance problems. Some of these problems may be managed in the community or primary care. However, balance problems can be complex, involving multiple clinical specialties, and so careful consideration needs to be given to the cost-effectiveness of these pathways and the best utilisation of resources. Survey respondents and focus group participants suggested that models of care to be considered should focus on the symptoms of the patient, not on single body systems or clinical specialties, with the role of multidisciplinary clinics being considered. An added complication is that little is taught on ENT and balance problems at medical school, with the result that directing patients to the right pathway from initial presentation may be challenging. The indications for referral to specialist and sub-specialist management need to be clarified and strategies for sharing this knowledge effectively need to be determined, in order to ensure that patients benefit.

This is related to the research priority in this topic where clear diagnostic criteria for different balance disorders need to be determined. These also require research to improve knowledge about the individual causes of dizziness. Diagnostic criteria based on this understanding has a clear benefit for patients: it lets them know what is wrong with them, what to do about it and gives them an idea of what to expect from their condition in the future. Moving forward there needs to be consideration of developing data collection platforms to collect the information needed for this research.

Patients noted that the impact of balance conditions on their lives was often unappreciated and its causes misunderstood. They felt that in future research a patient-centred approach that recognised the lived experience of these conditions was vital, and that the role of self-management and patient support needed to be investigated to determine what strategies had the greatest impact on their quality

of life and disease outcomes. Healthy physical and social environments, including designs of buildings and communities and educating policy-makers and the public about their importance, were ideas considered by our patient respondents, so that people with balance problems are not limited in their ability to interact with society.

The Priorities
- What are effective interventions for hearing loss in older people?
- Is it beneficial to introduce an adult hearing screening programme and how do we best develop it?
- What is the optimum design for "balance pathways" in older people with balance problems?
  - How and where can older people with balance problems and with multiple medical conditions and decreased mobility best be managed?
  - How can self-management strategies be built in these pathways and what is their impact on patient outcome?

The importance of effectively managing ENT, Hearing and Balance conditions in an ageing population was mentioned by many survey respondents and in the focus groups. Consideration needs to be given as to what is meant by an ageing population; there can be a marked difference between biological age and chronological age. When considering ageing and the older patient this difference gives rise to different considerations: the management of older people with multiple medical problems as opposed to the expectations and quality of life of the ageing but fit population. The expectation of either of these groups is that if you live long enough you will develop some degree of hearing loss or balance problems, and these have driven the chosen priorities.

Research into effective interventions for hearing loss have the potential to impact on large groups of people, making this idea one of the top research priorities in this topic area. It was recognised that there needs to be better understanding of why age-related hearing loss develops, as this could lead to interventions to reverse or stop it without resorting to hearing aids. Although the design of hearing aids has improved, older people in particular can find them especially difficult to use if they have poor hand function or struggle with new technologies, and there is concern that there is a stigma attached to their use. Respondents also considered that research on how to improve access to hearing services for older people would be useful. However, in the Consensus Conference it was felt that better devices and treatments for hearing loss, as well as healthy environment interventions, such as acoustically friendly buildings, would have an impact both on individual quality of life ("a real impact at the point of intervention") and on population health. It was recognised that intervention for hearing loss in older people should also involve family and carers.

The development of an adult hearing screening programme as a research priority is related to the development of effective devices, treatments and environmental interventions, as people could then benefit from early identification. The ability to screen for hearing loss and stop progression or restore hearing would "be a game changer." It would transform expectations of quality of life, and could play a huge role in vulnerable populations, such as those suffering from dementia, as well as maintaining cognitive health in older people. How best to screen for hearing loss and which tests to use for a screening programme needs further research. There are also questions around whether earlier diagnosis of hearing loss has an impact on patient

Reasons for these choices

Effective interventions for age-related hearing loss would impact on a large group of people, improving individual quality of life and population health.

These interventions for age-related hearing loss combined with the public health intervention of an adult hearing screening programme could transform expectations of quality of life and have a significant impact on vulnerable populations.

Balance pathways in older people can have an impact on quality of life and on the health burden of falls, and need to consider the potential for multiple medical problems.

experience and outcome, who should be screened, and the cost-effectiveness of screening. Screening was also seen as a good research tool to gather population data that can increase the understanding of hearing loss.

Research to identify the best pathways to manage balance problems both in fit older people, as well as in the vulnerable who may have decreased mobility, be house bound or in residential care, is also a key research priority. This has an impact on maintaining ongoing quality of life and reducing the health burden associated with falls. This is similar to one of the priorities in the Balance topic, but emphasis here was placed on the need to take a holistic approach to older people in consideration of their potential for multiple medical problems and the possibility they may be in residential care, and to include research into effective self-management strategies and their role in pathways.

Children and Young People

The Priorities

- What outcome measures and instruments should we use for common childhood ENT, Hearing and Balance conditions, based on our understanding of the impact of these conditions, for otherwise healthy children and for those with complex needs?
- How do we best diagnose and manage obstructive sleep apnoea (OSA) in children?
- How can we improve knowledge of childhood ENT conditions and their evidence-based management for healthcare professionals in different settings, and what is the impact of this on patient outcome?

From both the survey and discussions throughout the initiative it was felt that more research was needed about the impact of ENT, Hearing and Balance conditions on children, their families and society. There was a need to develop agreed outcome measures for future research of the management of these conditions that captures this impact. Current research measures outcomes that are too focused on numbers and the results of medical tests; more meaningful outcomes - in terms of patient and family experience and quality of life - need to be agreed by patients, clinicians from different disciplines and researchers. Many different people and professionals are involved in the care of children, and any research done should involve children and parents, and include multidisciplinary research teams.

Children with complex needs are often excluded from research into the effects of treatment of ENT, Hearing and Balance conditions, although the impact in this vulnerable group may be the greatest. Hence the importance in future research of considering *both* otherwise healthy children *and* those with complex needs was emphasised. The potential for inequality in accessing services for these patients, and for those from different ethnic and socioeconomic backgrounds, is recognised and needs to be considered in research that develops pathways for their care.

The following ENT, Hearing and Balance conditions were identified as either common, have a significant impact on children, or both: glue ear; recurrent ear infections; sore throats; coughs and colds; nasal obstruction and allergies; snoring and OSA. Congenital and acquired inner ear (sensorineural) hearing loss was also recognised as a condition requiring further research into the choice of best outcome measures and evidence-based management. Obstructive sleep apnoea (OSA) was singled out due to the level of concern about this condition from parents in terms of its impact on their child's development and school performance. It was felt that there is variation and uncertainty amongst clinicians in various settings about how to diagnose and treat it, and that research in this area has been under-resourced. The prioritised question about OSA specifically involved pathways of care and standardising diagnostic criteria.

Improving knowledge and education of ENT, Hearing and Balance conditions in children

Reasons for these choices

ENT, Hearing and Balance conditions are very common in children.

To improve the value of future research the impact of conditions on children and families needs to be better understood. Research outcomes need to be agreed upon by researchers, clinicians and parents so that research is patient-centred and results can be compared across studies.

OSA is of great concern to parents and can have a significant impact on children and their development and school achievement; there is uncertainty and variation in how it is best managed.

Knowledge of ENT, Hearing and Balance conditions is variable in different health care settings, despite the large burden of disease of these conditions.

in different settings, such as the community, primary and secondary care, and amongst different healthcare workers was important to patients and professionals. Future research should be developed in collaboration with parents and carers and include better recognition and self-management of these problems leading to improved outcomes for children, which links to the first priority in this topic.

Infection, Inflammation and Allergy in Adults

Rhinosinusitis is common and survey respondents noted that many uncertainties exist around its treatment for various subtypes of the condition. Thus broad questions were developed in order to include a range of specific research questions, with the focus removed from a single disease subtype. After discussion it was suggested that strategies should not only include treatments but also prevention, so that lifestyle or environmental strategies to prevent development of chronic ENT infections or inflammatory conditions were included.

The research priority 'define different variations of disease,' was felt important as it provided knowledge for improved diagnosis, allowing for 'personalised medicine' with treatments based on these specific diagnoses, risk factors and individual disease markers. This would speed up diagnosis of rhinosinusitis as both patients and healthcare professionals noted this may come too late, exacerbating conditions and causing distress to patients. They felt that this was due to a lack of basic understanding of conditions, inadequate sharing of the knowledge that we do have, and failure to appreciate the significant impact of the condition on quality of life. Thus the basic understanding of conditions was important. It was also recognised that agreed outcome measures relevant to patients and professionals were needed to inform future research, to ensure that it was of value and would have an impact on clinical practice. The combination of the ideas about defining disease, treatments and outcome measures was felt to be a good basis for a research agenda.

There was firm agreement that antibiotic misuse and its impact on antibiotic resistance is a major issue. It was noted that there are guidelines on management of ENT infections in primary and secondary care; these may need to be updated and more strategies to improve their implementation need to be developed.

Loss of smell and taste, and the importance of improving knowledge of its causes and measurements of function, were raised by patient survey respondents. Anosmia has a significant impact on quality of life, and this should be reflected in outcome measures of future research. A loss of sense of taste and smell were recognised as potential indicators of neurodegenerative disease, such as Alzheimer's and Parkinson's disease, and these associations would benefit from further investigation.

It was felt that there were two missing questions:

- Cell-based therapies for sensory ENT disorders; this was raised across all areas and will be incorporated in this topic too.
- Patient representatives were particularly interested in what patients can do to improve their own care and that developing information on this would be of benefit.

There were many questions raised in the survey about allergies such as seasonal and perennial allergic rhinitis, and in particular the role of immunotherapy. These have not been specifically included in the final agenda as colleagues in immunology and other allied medical specialists in collaboration with ENT and primary care have active research programmes already addressing these areas.

The Priorities

- What outcome measures and instruments should we use for research into head, neck and airway disease, both benign and malignant?
- What is the optimal management for adults with OSA?
- What is the optimal management for adults with voice problems?
- What is the role for improved diagnostics in head, neck and airway conditions?

Developing agreed outcome measures that are relevant for patients and clinicians was an important issue raised across the topic areas. The importance of understanding the impact of head, neck and airway problems on patients and determining what is meant by optimal management was highlighted. This discussion originally focused on benign head and neck conditions, but it was felt that this was relevant and important for head and neck cancer too. Agreeing on outcomes for future research increases its value as it allows study results to be compared and combined.

Obstructive sleep apnoea (OSA) in adults was highlighted as an important area for research. This affects a large proportion of the population and has a significant impact on health, quality of life and safety of individuals. It was felt that optimal management of OSA should include self-management and patient education, as this could have the added benefit of reducing the burden on secondary care, making the patient pathway more efficient and cost-effective. It was recognised that although treatments such as CPAP are available for OSA, these are of variable benefit for patients and that there is scope for improvement in the management of this condition. In recognition that patients with OSA often have comorbidities, it was felt that there has to be better communication and collaboration between disciplines both in the management of patients and in research about this condition, and to consider the patient as a whole.

The management of voice conditions was felt to be a significant area of unmet need, with patients uncertain as to what treatments were available to them and how to access them, and with variation in management and access to services across the country. Current management has been largely based on "received wisdom" and the evidence base needs to become more robust. Although this does not affect the same numbers as OSA, patients felt that the impact on individuals was significant. The voice was felt to be important to personal identity; conditions affecting the voice had a potential impact on employment, social interaction, and wellbeing.

Reasons for these choices

Agreed outcome measures will improve the value of research investment and ensure that the results are relevant to patients.
OSA was highlighted due to the large clinical burden of disease, and the potential to develop appropriate pathways that avoided inappropriate referral to secondary care.
Voice problems are a substantial area of unmet need for patients.
Improving diagnostics will have an impact on patient care, outcomes and quality of life.

Accurate diagnosis of head, neck and airway problems is the first step leading to appropriate management, which compliments the priorities that focus on appropriate management. Improved and more rapid diagnosis relieves the stress on patients and reduces the added health problems due to delay.

Further head and neck cancer questions were raised by survey respondents, but many of these were already the subject of research in progress by cancer researchers and therefore were not included in the final agenda.

Creating a Research Culture in ENT, Hearing and Balance: Towards a Strategic Research Agenda

Throughout the development of the research agenda, from the initial data gathering by survey and focus groups, to the in depth discussions from the Expert Forum and Consensus Conference, key ideas about what the final agenda should incorporate and strategies for implementing it emerged. Together they form the seventh theme of the framework, 'Developing a Research Culture in ENT, Hearing and Balance' and will inform the next steps after the launch of this agenda.

Patients and professionals wanted this agenda to contain:

Patient-Centred Research and Self-Management: Putting patients at the centre of research was seen as key to this agenda. Patients and carers have been central to developing this agenda; they have been involved in every step of the process. Ongoing collaboration with patients and carers to deliver the agenda and develop the research projects will ensure that future research in ENT, Hearing and Balance is patient centred. Patients voiced the importance of better communication and sharing knowledge between healthcare professionals and with patients in developing patient pathways for common ENT, Hearing and Balance conditions. Patients particularly wanted research that would include self-management and peer-support as part of management strategies.

Research Leading to Sustainable Health Care: Ensuring that research leads to the development of sustainable high value health care in ENT, Hearing and Balance in a system that is recognised as stretched, with growing demand placed on it, was seen as an important goal throughout this initiative. Sustainable health care depends on improving the overall health and wellbeing of the population, and encouraging people to take responsibility for and manage their own health as well as developing efficient, cost-effective and high value services.

Novel Therapies: The development of cell-based and molecular therapies across ENT, Hearing and Balance conditions was seen as a long term but important research priority by both patients and professionals. It was recognised that it would take time for these novel therapies to find their way into clinical practice and provide benefit for patients; these long term strategies were considered an important balance to some of the shorter term research priorities.

In order to implement this agenda, consideration needs to be given to the following:

Capability and Capacity: To implement this research agenda and grow the workforce to develop and deliver the projects, there needs to be understanding of current capability and capacity for research in ENT, Hearing and Balance. Mapping current research activity across the translational pathway - who is doing what research and with whom do they collaborate - is necessary to see where further growth and development is needed. Close collaboration with the NIHR Clinical Research Network and its ENT group will be key to the planning of research carried out in the NHS. Importantly, the resources needed to support the advancement of clinical academics in ENT, Hearing and Balance needs to be mapped against what is currently available in order to identify them and the areas needing further development. Future researchers will need guidance and mentoring; the regional ENT trainee research collaboratives and Interact, their recently established national working group supported by ENT-UK, can play an important role in this process.

Collaboration: Patients with ENT, Hearing and Balance problems present to multiple health care disciplines and across a variety of health care settings. In order to develop and deliver the high value translational research that has the potential to change practice, research collaborations need to be formed across both health care and academic disciplines. Key clinical collaborations considered were across community, primary and secondary care through a variety of specialties including GPs, Physiotherapists, Paediatricians and those caring for the elderly. Key academic collaborations discussed were with Health Informatics, Innovation and Implementation Science and Health Services research. The need for expert qualitative input into the traditionally quantitative field of ENT, Hearing and Balance research was felt to be important. So was seeking collaborations to help span the gaps along the research pathway, improving translation of research from bench to bedside. This approach will facilitate the breakdown of the traditional silos of research and clinical care to develop knowledge and services that are sustainable in modern health care.

Developing Data Collection Platforms: Embedding research in day-to-day clinical practice and collecting and using "real world health data" to develop knowledge that is relevant to patients and professionals was raised throughout the initiative. This was described as developing electronic health records that capture information that is both applicable to clinical care as well as to research, and as developing databases or registries of specific ENT, Hearing and Balance conditions and interventions; this is the concept of developing data collection platforms. These will require significant resources and collaboration with the operational side of the NHS to integrate them into existing and future IT systems.

Outcome Measures: The need to gain a better understanding of the impact of ENT, Hearing and Balance conditions on patients and their families was raised across the whole spectrum of the research themes and topics. It was felt important that clinicians and researchers should work with patients and carers to decide which outcomes to include in future research. The development of so-called 'core outcome sets' across a range of conditions and interventions requires a strategic approach with patient and professional organisations deciding which conditions to prioritise and how to ensure that they are kept up to date. Careful thought needs to be given as to how best to secure and effectively use the financial and human resources to do this. One of the benefits of this being a strategic priority is that it can give rapid return on investment as these 'core outcome sets' can be developed in a relatively short time frame and have wide applicability.

The Team

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