Advancing the science of outcome measurement in paediatric palliative care: views from multidisciplinary healthcare professionals and advocates

Running title: Paediatric outcome measurement

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Conflict of interest

The authors state that they have no conflict of interest.

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Reflective questions:

1. Why do outcome measures matter in improving care?
2. What are the specific challenges to outcome measurement for children, young people and their families living with life-limiting and life-threatening conditions?
3. What are the key features of a feasible and acceptable outcome measure?
4. How would you go about implementing an outcome measure in your practise?

Abstract

Background

We lack appropriate person-centred outcome measurement (PCOM) to direct and evaluate care.

Methods

In line with COSMIN measure construction guidance, we convened an expert group to elicit views on 1) domains/items to include; 2) implementation challenges; 3) requirements for use in routine care by practitioners. Data were content analysed.

Results

N=36 UK-wide clinicians, advocates, and researchers participated.

1) Items for inclusion were: specific symptoms, education, play and social interaction, parental time for partner & other children, sex & intimacy, and sibling wellbeing. 2) Implementation challenges: supporting CYP to engage meaningfully, the instrument being seen as a “test” of parents’ care quality, raising unrealistic expectations, proxy validity. 3) Need for clear administration and interpretation guidance, agreeing data ownership/access.

Conclusions

This expert meeting addressed the initial step in COSMIN guidance, informing face validity and acceptability. It provides the information necessary for the first phase of tool development and informs potential utility and implementation.

Background

Longer survival for children and young people (CYP) with complex incurable conditions has led to trajectories with slow deterioration, high dependency and disability.(Watson, 2009) 2001-2011 data show rising prevalence of life-limiting childhood conditions in England (from 25 to 32 per 10,000 population), with increases greatest among non-white and deprived regions(Fraser et al., 2012). There were 206,505 English hospital admissions of CYP with life-limiting conditions (LLC) and life-threatening illness (LTI) in 2011; congenital abnormalities, neurological, oncological, and respiratory were the four most common conditions from the International Statistical Classification of Diseases and Related Health Problems (ICD-10) (Fraser, 2011).

Palliative care needs and care provision for CYP and their parents differ greatly from those of adults. Differences include the need to be aware of for the child's developmental phase as well as their chronological age, language and communication needs, earlier intervention and longer unpredictable trajectories, social and educational engagement, play needs, and parent support needs (Paediatrics, 2007),(Lives). In common with older adults, CYP are often dependent on families for care, and there may be complex issues with respect to clinical, legal and ethical decision-making. These potential differences need to be recognized in both the development and implementation of person-centred outcome measures (PCOM) for CYP.

Very little research has been undertaken on palliative care outcomes for CYP with LLC and LTI. Evidence in the UK has shown that referral to specialist paediatric palliative services reduces subsequent hospital admissions for children with cancer compared with those not referred(Fraser et al., 2013). It is important to be able to also measure person-centred outcomes across the range of providers and care settings. Most studies to date have used qualitative, retrospective and proxy methods(Drake et al., 2003, Wolfe et al., 2000), i.e. there is a lack of prospective self-report data that quantifies outcomes. While existing data has provided some insight into the concerns of CYP with LLC and LTI (Namisango et al., 2018), the evidence is not sufficient for development of PCOMs that are both valid and reliable.

PCOMs can improve the quality and equity of care (Dawson et al., 2010). A PCOM for CYP and parents receiving palliative care would firstly better enable them to identify their priorities and outcomes of care with their clinicians. Second, their clinicians would be able to conduct more thorough assessment and monitor the outcomes in their care planning. Third, commissioners would be able to ensure that the commissioning for these specialised services is “patient-centred and outcome based” in line with UK specialised health service guidance.

Adult measures applied to CYP lack content and face validity. This is due to the absence of appropriate concepts and language (especially around learning, play and social activities) and key elements of child wellbeing(Ravens-Sieberer et al., 2006). A review of paediatric palliative care research identified as a priority the development of a palliative care outcome measure specifically for CYP . The review also noted that appropriate scientific methods must be developed to ensure that PCOMs are appropriately developed and implemented for this population(Ullrich and Morrison, 2013). The development of outcome measurement in the two fields of CYP health and palliative care are UK research priorities.(MRC, 2009)

Tool development for this population must begin with careful consideration of the conceptual basis of any proposed measure(Oxford, 2009). A validated tool for CYP receiving palliative care is the Memorial Symptom Assessment Scale (MSAS), a 32-item symptom inventory that measures pain and symptoms with children with cancer(Collins et al., 2002, Collins et al., 2000). This validated tool is appropriate for measuring pain and symptoms, but as such addresses only one dimension of palliative care. Further, the tool may be too lengthy to be of use in routine clinical practice and not well suited for CYP with non-malignant disease, communication and/or learning difficulties, who constitute a major portion of the population of CYP with LLC and LTI(Bausewein et al., 2011c).

Similar problems exist with the PEDSQoL (a commonly used quality of life tool for CYP). A study in children’s palliative care concluded that it lacks validity due to its physical function focus (Huang et al., 2010). A recent systematic review of outcome measures in paediatric palliative care examined measurement properties of 27 instruments, concluding that the domains, as well as recall and response format, were not considered appropriate for paediatric palliative care populations(Coombes et al., 2016). A paediatric version of the Palliative Outcome Scale (POS) has been developed for CYP in sub-Saharan Africa (Downing et al., 2012), and is seen as a “milestone” in the field(Downing et al., 2018).

This responds to deficiencies in currently available measures, but may not have face, content or construct validity for outcomes in a high income country setting.

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PCOMs for CYP receiving support from palliative care services, and the methods to ensure participation in measurement, are urgently needed(Knapp and Madden, 2010, Harding et al., 2017). There are escalating calls for outcome measures to support paediatric palliative care research, and to assist in the clinical goal of reducing suffering (Sirkiä et al., 1997, Hechler et al., 2008, Contro et al., 2002, Wolfe et al., 2000). In this paper we report expert views of researchers, clinicians, and advocates on the utility and conceptual properties that would underpin development and validation of a PCOM for CYP with LLC and LTI and their parents/caregivers.

Methods

Design

In line with guidance on the development of measures(Kline, 2015), we invited experts (clinicians, researchers, advocates of paediatric palliative care from across the UK) to participate in a whole day consultation to inform development of a PCOM in paediatric palliative care (i.e. CYP facing LLC and LTI up the age of 18). Engagement of health professionals at early planning stages can build their understanding of the rationale and usefulness of a PCOM (Boyce et al., 2014).The meeting aimed to bring together this group of expert stakeholders as the first step in informing development of a PCOM with optimal potential to fit into existing practice, clinician decision making, and healthcare organisation(Greenhalgh, 2009).

Objectives

The objectives were to 1) elicit potential domains/items to be included in a person-centred measure for CYP with life-limiting/threatening conditions; 2) identify challenges in implementing a PCOM in this population; 3) identify requirements for the tool to be used by practitioners as part of routine clinical care. These were asked in the context of all LLC and LTI faced by CYP. Given the wide range of relevant conditions with potentially low prevalence (Hain et al., 2013), and outcome measure user views in palliative care that we should use a minimal number of tools (Bausewein et al., 2011b, Bausewein et al., 2011a), the discussion aimed to support a single core measure.

Procedure

An invitation was sent out to all clinical professionals and advocates from the membership mailing list of Together for Short Lives, the UK's national children's palliative care charity that supports a membership of hospice, hospital and community-based children's palliative care services as well as a community of young people and families. Each facility was asked to send a single delegate, and all those who accepted were provided with a place at the meetong. In addition, we invited experts in PCOM psychometrics and implementation in adult palliative care and researchers in special education who have conducted studies of CYP with communication difficulties(Dockrell and Lyndsay, 2011). In line with COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) guidance, we designated participants as "experts" in tool construction as "someone who possesses relevant knowledge, experience regarding the construct to be measured (e.g. clinical experience, published and respected by others)"(Kline, 2015).

Prior to expert discussion, the meeting began with a series of presentations. i) The theory and practice of PCOMs in palliative care, drawing on the development, implementation(Antunes et al., 2014), quality improvement (Etkind et al., 2015) and research programmes in adult care(Collins et al., 2015) using the POS (www.pos-pal.org). ii) Findings from a systematic review to appraise the validity and appropriateness of existing PCOMs with respect to palliative care outcomes of CYP(Coombes et al., 2016). iii) Conclusions from a UK working group on outcome measures for CYP with LLC and LTI, which identified the need for clinical and academic partnership to develop and new measure(2015) . iv) Development and validation of the African C-POS(Downing et al., 2012). v) Development of a UK goal attainment scale for CYP attending palliative care (Harris, unpublished).

Following the presentations, participants were allocated to three discussion groups (one for each objective) to achieve a mix of professions. Each group was asked to respond to one of the three objectives with a dedicated scribe. Following discussion, each group fed back to the full expert group. During feedback, additional responses to each objective from the full group discussions were also recorded. The written notes of expert opinions resulting from each discussion group were content analysed to address each of the aforementioned objectives. This analysis was conducted initially by the first author (RH), and the proposed groupings developed from the workgroup notes was reviewed and the findings agreed with the remaining authors (MBL and LC). We did not seek consensus at this initial stage of tool development but aimed to identify the range of views. Findings from the expert discussions are reported by objective below. We did not seek ethical approval for this consultation exercise and do not use direct quotations.

Results

Participant characteristics

There were 36 participants from England, Wales, Scotland and the Republic of Ireland from a range of disciplines and professions. These included: paediatric palliative care nursing (n=15), paediatric palliative care medicine (n=10), advocacy (n=3), research (palliative care outcome measurement and tool construction n=3, paediatric palliative care social scientist n=1, epidemiology n=1, and special education n=1), law (n=1) and service user representation (n=1).

Findings

The participants participated fully in discussion. Responses to the questions are reported by objective and summarised in Table 1.

Objective 1: Potential domains/items to be included in a PCOM for CYP with LLC and LTI.

Participants endorsed the multidimensional WHO definition of palliative care, and agreed the PCOM should measure physical, psychological, social and spiritual outcomes. For CYP, the expert members identified social concerns to be considered for a PCOM, including education, play, social interaction and sibling welfare. A number of specific symptoms were also proposed, including seizures. The proposed parent outcomes were specific to CYP palliative care, and included time for partner & other children, sex & intimacy. The issue of sibling wellbeing was strongly underlined throughout the meeting and should be considered in addition to the target PCOM users of CYP and parents.

Objective 2: Challenges in implementing a PCOM in this population.

Participants identified some key purposes and features of a PCOM and the challenges that these may bring. These included ensuring that the instrument could be used by CYP with a wide range of cognitive abilities, is applicable throughout the child's life and into end-of-life, that the outcome measure not be seen as a "test" of the quality of informal parent care provision, that the instrument be "child-friendly" engaging and brief, and that there be appropriate mechanisms so that individual results could be shared across the multiple agencies and services involved in care. Although the PCOM should have demonstrable proxy validity, parents' outcomes are important in their own right, and parents should not be seen as solely proxies for CYP. The expert group expressed conflicting views about whether sibling outcomes should be measured in PCOM as had been suggested for Objective 1.

For Objective 3: Implementing the tool in routine care

Participants felt that the way the tool is perceived would be critical for implementation. They cautioned that while the tool must have established responsiveness, it must avoid raising unrealistic expectations of care. The purpose of outcome measurement should be clearly aligned to improving person-centred care, and not an instrument with a solely "political" (i.e. funding) purpose. They also noted that clinical teams should be trained in the appropriate interpretation of the results at an individual and population level. Finally, they noted the need for clarity on who would "own" and have access to data with clear guidelines on storage, access and use of the data.

Discussion

The presentations and discussions identified critical issues in design and implementation of PCOM in CYP with LLC and LTI. The findings of the expert discussion groups established some important common ground. First, among the expert participants there was recognition of a need to develop outcome measurement for CYP. Second, there was a real desire to move forward to develop the measure. Third, there are many challenges that will need to be addressed, including reaching agreement on which items are to be included, how best to promote routine use among stakeholders, and to how the data should be interpreted and utilised. With respect to this third point, the expert meeting provided important information for the initial stages of developing a valid PCOM for CYP and their families receiving support from children's palliative care services, with adequate face validity, and acceptability. (Mokkink et al., 2010)

This expert consultation and discussion has been an essential first step in the development of a valid PCOM. Given the lack of service user views, we will now undertake detailed qualitative investigation with CYP, parents, siblings and health care professionals to inform item generation. The exchange of views during expert discussion addressed the need to engage and establish views of health professionals on content, format and usage at planning stages(Boyce et al., 2014)·(Antunes et al., 2014). Early involvement of stakeholders is essential given the evidence that although PCOMs improve discussion and detection of problems, their impact on clinical management depends on how "*PCOMs fit (or do not fit) into the routine ways in which patients and clinicians communicate with each other, how clinicians make decisions, and how healthcare as a whole is organised*"(Greenhalgh, 2009). In terms of identifying appropriate PCOM formats and methods of engagement, PCOMs that have been developed and validated in fields such as children's disability may be useful. Existing PCOMs for children with communication difficulties may offer useful mechanisms of engagement in person-centred measurement with self-report to ensure that end users have optimal opportunity to participate.

Given the relative infancy of outcome measurement in paediatric palliative care, the lack of prior consensus on generating and implementing a valid tool with maximum utility, and the paucity of available data to inform a measure(Downing et al., 2018), this consultation enables us to move to rigorous too development and validation. In line with COSMIN guidance, the findings from this consultation will inform the face validity of this tool, although the "expert" view to inform content validity must be that of the CYP and their family. We believe that a valid PCOM will also support current initiatives such as "Getting it Right for Every Child" and the "Anticipatory Care Plan for Children".

This is the first report of stakeholder views on PCOM properties and usage for CYP in palliative care. A limitation is that we do not yet have views directly from CYP. This will be conducted as the next phase of item development, with data collection from CYP and their families to determine content validity. A further potential limitation is that although we were able to recruit multidisciplinary experts from across countries within the UK, priorities for items and face validity may vary according to geographic region. However, cross-national study has identified common interpretable items for adult palliative PCOM content.(Schildmann et al., 2016) Lastly this study did not seek to use qualitative methods generating direct quotations, but to generate a summary of the breadth of discussion. The detailed qualitative work required to establish face and content validity is now being designed, with subsequent testing of reliability.

We have now established stakeholder support for the principles of PCOMs in this population to inform initial face validity, acceptability and use in routine practice. This enables us to proceed to tool development and validation in line with current best practice (Mokkink et al., 2010) in health outcome measurement.

Author's contributions

RH conceived and led this study, MBL and LC assisted in design, interpretation and manuscript drafting. All authors approved the final manuscript.

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Author Disclosure Statement

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Table 1 Respondent views on a proposed palliative care outcome measure for children and young people

Obj 1 Outcomes to be measured		**Obj 2 Specific implementation considerations**		**Obj 3 Considerations for use in routine care**	
i) Should address 4 domains of the WHO palliative care definition	-Avoid a functional focus -Include physical, psychological, social spiritual outcomes	i) Format	-Usable for range of cognitive abilities -Relevant for palliative *and* end-of-life care -Avoid “testing” parent care -Reflect culturally diverse users -“Child friendly” and engaging -Avoid jargon in tool name -Brief length	i) Functions PCOM should perform	-Be able to evaluate services -Detect changes under care -Avoid raising unrealistic expectations -Be person-centred and not a tool to meet political ends -Avoid only capturing “negative” -Be clinically relevant -Work across involved teams
ii) Specific items	*Specific symptoms:* Distress/ agitation/ pain/ nausea/ vomiting/ secretions/ breathlessness/ skin integrity/ seizures/ constipation/ secretions/ fatigue /respiratory	ii) Population	-Should address all ages including antenatal and transitions to adult care, development stages, language development, conditions -Danger of being a measure that plots deterioration	ii) Ease of interpretation	-Change management -Capacity in small organisations -Attribution of change to care -Responding with ‘right people, right place, right time’ -Easy visual interpretation

	Parent: Time for partner & other children/ respite/ sex & intimacy/ sleep/ engagement in activities/ “Can you face the day”?/ confidence to care/ decision making involvement/ care involvement *Psychological:* Empowerment/ sharing feelings/ sadness/ feeling overwhelmed/ coping/ anxiety/ acceptance *Care processes:* Information /communication/ coordination/ end-of-life &			-Many agencies and services are involved, need to establish MDT working in response to scores -Require “trust” to be established with the participant			

	place of death planning/ parent involvement *Social concerns:* Employment/ isolation /education/ play/ social interaction/ sibling welfare/ independent living/ financial security/ sexuality/ housing /school attendance/ child engagement in activities *Spiritual wellbeing:* Being at peace						
iii) Use open items to identify what matters most			iii) Completion	-Communication with nonverbal children, cognitive abilities and use of communication aids -Distinguish between parents as proxy vs reporting own outcomes		iii) Data usage	-Know whom to contact & respond to need at any time -Demonstrate PCOM benefits to parents -Clarity on data ownership & access -Analysis guidance

to parents and children				-Determine proxy validity -Risk of parent agenda influencing child self-report -Ethical challenges of talking to children without parents -Determine optimal recall period -When to introduce the tool			-Ethical guidance on data collection, usage, storage -Establish a central repository

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