

Managing and sharing research data in children's palliative care: risks, benefits, and imponderables

1. Introduction

Children's palliative care (CPC) is a growing specialist area, focussing on holistic support for children and young people with life-limiting illness. This encompasses symptom control for the child, emotional and psychological support for the child and family, and addresses practical, financial and spiritual needs. Support from professionals may be required over an extended period from the time of diagnosis of a life-limiting condition, continuing throughout the child's life, and extending to include bereavement support for surviving family members after a child or young person has died.

When the speciality was in its infancy learning was based on experience and sharing good practice, but there is now an increasing emphasis on evidence-based interventions and care delivery, requiring the development of a robust and rigorous approach to research which will stand up to the highest levels of scrutiny (Lioffi et al, 2017; Baker et al, 2015). There is an expectation that during academic studies, publication of peer-reviewed papers or production of reports to funding bodies, this scrutiny will include access to the data collected as part of the research process (Concordat Working Group, 2016). Access to research data by those outside the research team raises some important issues for research in this area, amplified by the challenges of research with a small population. In this editorial we explore what this means for those conducting research, and for participants in research. We then highlight areas for further consideration to inform a conscientious approach in the future.

2. Research Data Repositories

In health and social sciences electronic data capture and storage media have facilitated the collection of large amounts of information given by individual research participants.

Research data repositories (RDRs) are data storage centres where research data can be submitted, stored, scrutinised, and subsequently accessed by researchers for purposes beyond the original intent (ie secondary uses; Manhas et al., 2015; Karcher, Kirilova & Weber, 2016). The use of RDRs is widely encouraged and frequently required by academic institutions, publishers, funders, and national directives such as the Medical Research Council, National Institute of Health, and the Social Sciences and Humanities Research Council 2012 (Concordat Working Group, 2016; Bishop & Kuula-Luumi, 2017; Van den Eynden et al., 2011). Access to research data by researchers outside the original research team can elevate the impact, efficiency and effectiveness of scientific activities and funding opportunities (Crane D., 2018; Van den Eynden et al., 2011; Corti & Thompson, 2006; Lee & Stvilia, 2017). RDRs give many crucial benefits, including the opportunity to scrutinise and verify research findings; improving the transparency of research procedures; ensuring the maximum benefit is derived from research data sets via access to “unmined” data; avoiding duplication; reducing the burden on research participants; creating opportunities to re-examine data with new perspectives; and the recognition that the output of publicly funded research is a public asset (Bishop, 2009; Sherif, 2018). However, these advantages must be balanced against both the need to comply with the legal framework concerning data protection regulation and the ethical requirements on researchers to protect the privacy of the participants in their studies (Bishop, 2014).

The issues regarding secondary access to research data apply to both quantitative and qualitative material (Antman, 2014). Particular concerns have been raised about data storage, access, and re-use of sensitive or personal data (Manhas et al., 2015; Lee & Stvilia, 2017). Children’s palliative care research is an area where these issues are particularly poignant for research participants given that the research data may include contextual information about individuals, their families, health and relationships that could potentially identify them.

Qualitative data may also include reflections on personal decisions and value judgements at a very stressful time, when participants may be feeling overwhelmed and vulnerable.

3. Potential Approaches to the use of RDRs in Children's Palliative Care

There are three strategies recommended to safeguard research participants' research data stored in RDRs whilst maximising the potential benefit from it: effective anonymisation of data to prevent breach of confidentiality; obtaining consent specifically for the long-term storage and potential re-use of data; and control of access to the data once stored in the repository (Morrow, Boddy & Lamb, 2014). All three present challenges for researchers in CPC.

Anonymisation of data can be difficult in the small world of children's palliative care.

Children with rare conditions or unusual social circumstances may be identifiable from minimal amounts of contextual detail (Manhas et al., 2016). Current MRC guidance recommends that under these circumstances research can continue but is subject to General Data Protection Regulation (GDPR) considerations, by managing data disclosure through consent (MRC, 2019). Individual clinical scenarios may identify not only the patient and family, but staff teams involved in providing support, particularly if the readership of research outputs is a relatively small and interconnected group. However, removing all contextual information may reduce the value of the data by itself, thus diminishing the benefits of review or re-use of the original data (Yardley et al., 2014; Parry & Mauthner, 2004). This area has been explored in the context of qualitative research in social sciences (Bishop, 2009), but there is no evidence about the re-use of data without contextual information in CPC.

Informed consent is an essential component of responsible research practice, but whilst the primary researcher may be able to seek consent for their own study, it is impossible to predict

how data may be used for research in the future. Individual participants (or their parent/guardians) may feel willing to consent to a study where safeguards can be assured, and a personal relationship with the researcher or research team is possible (Yardley et al., 2014). Some may feel a sense of altruism and seek maximal benefit from their research contributions by agreeing that these may be used for future unspecified research or educational purposes. Others may be reluctant to have their data considered by strangers, or from another perspective, and seeking permission for this risks loss of trust between the researcher and potential study participant. This could jeopardise research recruitment overall, which would be to the detriment of the drive to improve the evidence base for practice that RDRs are aiming to improve.

Requests for consent for secondary use of research data some time after the initial study may be difficult for other researchers who have no direct relationship with the participants and may be impossible if the dataset was anonymised. Consent that was previously granted by a parent with assent by a child may no longer be valid once that child has reached the age of consent themselves. In addition, in the field of CPC the children or young people may now be deceased, and approaches to their bereaved families will need to be handled with sensitivity. The question of “ownership” of data, particularly once participants have died, is complex (Parry & Mauthner, 2004) and the duty of medical confidentiality continues even if data protection laws do not apply.

Stringent control of access to data repositories is the third approach, either limiting this to *bona fide* researchers who will need to meet certain criteria and be accountable for their use of data, or by limiting what data may be accessible (eg metadata only), or by limiting access in time such as having an embargo in place for a specified period (Antes et al., 2018). These arrangements will need enforceable license agreements in place to specify how data will be used, that it will not be disseminated further, and that confidentiality will be respected. As

yet there is no consensus on how these decisions are made, enforced, and who is accountable for the appropriate use of RDR data, or for its destruction once GDPR permissions have lapsed. There is also little information available to explain these issues for lay members of the public as part of a consent process (Manhas et al., 2015; Lee & Stvilia, 2017).

4. Views of Research Study Participants

Although these potential solutions may all play a part in addressing the challenges of RDR use in CPC, there remains a lack of research evidence about the views of children, young adults, and their parents/carers about RDRs and data sharing.

There is limited information about the views of study participants about secondary use of their data in other settings. In Finland, a study examined study participants' views on the secondary use of 168 qualitative interview transcripts about equality, gender and life experiences and to their surprise found that 98% agreed to archive their findings for future use, despite these including sensitive and personal stories. The reasons given included a desire for the advancement of science and re-examination of the original research conclusions. Many perceived the re-use of their interview data as self-evident, rather than perceiving the interview relationship as private or secret (Kuula, 2010).

Seymour has conducted research with the elderly into their views about technology and natural death (Seymour, 2003), and with healthcare professionals and bereaved families involved in a case study series about the care of those who received continuous sedation until death (Seymour et al., 2015). She has reflected on her experience of seeking consent from study participants about archiving and sharing data. Older patients from the first study were generally enthused by the idea that what they were offering the study would continue to be of use for other researchers. The second project involved gaining consent for data sharing from patients and healthcare professionals who participated in interviews about continuous

sedation. Seymour found that attitudes differed, with nurses being more reluctant to share their interview data than the medical staff. Professionals working in hospices or community settings were more likely to consent to data sharing than those working in hospital.

Manhas (2016) explored parental perspectives on paediatric RDRs for non-biological data in Canada, and found that parents strongly supported the sharing of their own, and their child's non-biological data, but they did express reservations relating to privacy, relationships and governance that should be considered in RDR development. The authors noted that many of the parents found the concepts of data sharing and data repositories a complex topic, meaning that a great deal of information and time was needed for participants to provide truly informed comment. This lack of appropriate information or explanatory material should be addressed to enable a fully informed debate in future.

None of these scenarios can be directly equated to the situations faced by families facing the death of their child. Reports suggest that young people may see their contribution to research as part of their legacy, and entrust researchers to preserve this and maximise any benefit to others, but this aspect has not been explored in a systematic way (Earle & Blackburn, in press; Blackburn, 2018).

Research in this field may involve discussions with healthcare professionals and there is little evidence about their views on secondary use of their interview data, particularly where this includes reflections about sensitive or difficult aspects of their roles. In an era of increasing litigation and unconstrained use of social media, the preservation and potential secondary use of interview transcripts from healthcare professionals may be viewed as a threat and deter their participation in qualitative research.

Although there are many potential challenges, these must be viewed within a framework of improving the evidence-base for services supporting children, young people and families with

palliative care needs. The lack of high-quality research evidence in this field was notable during the development of the NICE Guideline for End of Life care for Children and Young People (NICE, 2016). Researchers have a duty of care to study participants to minimise harm, provide information to seek informed consent, and protect confidentiality. Researchers also have a responsibility to their colleagues in the academic community to maintain professional standards of conduct with transparency and integrity. And finally, in the wider public sense, there is a duty to produce quality research of wider social value, particularly when this involves the use of public funds (Bishop, 2009).

5. Areas for further development

At a national level there should be agreed guidance about the approach to data management and data re-use which reflects the particular issues for this sensitive area of research, acknowledges the legal framework for data protection, and accommodates the concerns of funders, publishers, research institutions, national patient groups and healthcare providers.

Secondly, the CPC research community should develop clear information about research data repositories and develop templates for consent documents to clarify the issues for research participants. These should be reviewed regularly to reflect current legislation and written for age-appropriate audiences in lay language.

And last but by no means least, it will be crucial to involve children, young people and their families in this debate and explore the extent and nature of any concerns from their perspective. Families who have experienced the trauma of facing serious illness or the death of their child may be highly motivated to help others learn from their experiences, and their contribution to research is invaluable. Children and young people may have views that do not concur with those of their parents, and these should be recognised in the debate. Their

perspective on the best approach to the storage and potential re-use of data will reinforce the development of a collaborative research culture based on values of trust and respect.

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