

Manning J, Hemingway P, **Redsell SA**. Survived -so what?: Identifying priorities for research with children and families post-PICU *Nursing in Critical Care*, May 2017, doi: 10.1111/nicc.12298

Abstract

Background: The involvement of patients and the public in the development, implementation and evaluation of healthcare services and research is recognised to have tangible benefits in relation to effectiveness and credibility. However, despite >96% of children and young people (CYP) surviving critical illness or injury, there is a paucity of published reports demonstrating their contribution to informing the priorities for aftercare services and outcomes research.

Aim: we aimed to identify the service and research priorities for childhood critical illness survivors with CYP, their families and other stakeholders.

Design/Methods: We conducted a face-to-face multiple-stakeholder consultation event, held in the Midlands (UK), to provide opportunities for the experiences, views and priorities to be elicited. Data were gathered using write/draw and tell and focus group approaches. An inductive content analytical approach was used to categorise and conceptualise feedback. A total of 26 individuals attended the consultation exercise that included: CYP critical care survivors; their siblings; parents and carers; health professionals; academics; commissioners and service managers.

Findings: Consultation findings indicated that future services, interventions and research must be holistic and family centred. CYP advisors reported priorities that focused on longer term outcomes, whereas adult advisors identified priorities that mapped against the process of survival from the PICU. Specific priorities included intervention development and testing to address unmet communication and information needs throughout the pathway of recovery;

support and enhance the lives of PICU survivors including initiatives to empower families to self; support longer-term functional and psycho-social outcomes.

Conclusion/Relevance to clinical practice: This consultation exercise provides further evidence as to the value of meaningful PPI in identifying the priorities for research and services for PICU survivors and illuminates differences in proposed priorities between CYP and adult advisors.

Introduction

Globally, there is burgeoning demand for patient voices to be central in setting the focus and development of healthcare services and research (Ocloo and Matthews, 2016, World Health Organisation, 2008, Coulter and Ellins, 2006). Furthermore, the NHS Constitution (Department of Health, 2013), asserts it is imperative that the experiences of stakeholders, such as CYP and their parents, are incorporated into research studies to improve the accessibility and quality of healthcare. Subsequently, meaningful patient and public involvement (PPI) has become essential to the development, design and in some cases delivery of applied health research (Hewlett et al., 2006). However, one such population where there is a deficit in PPI are from those children and young people (CYP) that are critically ill and require admission, treatment and aftercare from the paediatric intensive care unit (PICU) (Tume et al., 2015, Menzies et al., 2016). Despite research participation by families with CYP who have life threatening conditions being shown to have a positive effects on participants, even when examining highly emotive areas (Steele et al., 2014), involvement in the development and design of PICU research remains limited (Menzies et al., 2016).

National statistics from industrialised nations indicate the overwhelming majority of infants, CYP survive a critical illness or injury that requires care from the PICU (PICANet, 2015). Therefore over the past decade there has been increased interest in the physical, functional, and psycho-social outcomes of this patient group and the development and testing of interventions to support those that survive (Manning et al., 2016, Ong et al., 2016, Rennick et al., 2014). The James Lind Alliance Priority Setting Partnerships (2014) names supporting patients and families post intensive care within its top three priorities, with priority question two stating, ‘How can patients and their families be best supported as they start living at home again (e.g. health and social care services, ICU support groups, long term follow-up)?’ However, this valuable priority setting exercise did not include the views of CYP or focus on PICU priorities. Furthermore, no published data exists from PPI activities that outlines the views and priorities of CYP and families that have survived PICU to inform the outcomes and aftercare research agenda. Subsequently, work is still required to elicit the priorities of CYP PICU survivors and their families to direct future health services and research.

The purpose of this paper is to report the findings from a multiple stakeholder consultation event that aimed to identify the priorities for services and research with CYP and families that had survived childhood critical illness.

Methods

Approach

The Royal College of Paediatrics and Child Health (RCPCH) acknowledge the importance of involving children and young people (CYP) in research. Subsequently, the RCPCH (2016) have developed an Infants’, Children’s and Young People’s Child Health Research Charter of Good Practice Principles which provides guidance for ethics committees to help them

scrutinise applications for studies involving CYP. However, in comparison to a more established PPI movement in the adult arena there is varying guidance, with an unclear evidence base, on how to do this with CYP and their families (Cavet and Sloper, 2004). The renowned organisation INVOLVE (2012), provide guidance about how to embed PPI within the research cycle, but this has been designed with adults in mind. Nevertheless we used the underpinning ethos of this approach to inform the PPI activities in this consultation exercise. Furthermore, we recognised that CYPs participation is active and nuanced in terms of equity and ethical issues inherent in paediatric research engagement (Clavering and McLaughlin, 2010). Therefore the participatory processes adopted needed to respect the child or young person's social relationships in order to promote their personal learning (Åkerström and Brunnberg, 2013).

The facilitators had previously undertaken exploratory research '████████████████████' with CYP and in line with the protocol (████████████████████) planned to disseminate the findings to participants and other stakeholders. However, we wanted to ensure the next phase of this research was built from the perspective of CYP, their parents and clinical staff. The original study was qualitative in nature therefore the “implementing” and “evaluating impact” aspects of the INVOLVE (2012) cycle are not relevant. The next phase focuses on identifying and prioritising future research for CYP PICU survivors. Therefore, a face-to-face consultation event to provide opportunities for the experiences and views of a range of relevant stakeholders to be gathered (Sequeira and Warner, 2007) was conducted in November 2015 in the midlands region of England, UK.

Setting and sample

In order to address limitations with the existing literature outlined by Menzies et al. (2016), we sought to consult and elicit feedback from a range of stakeholders that included: CYP that

had experienced a critical illness/injury; their parents and carers; siblings and other family members; health professionals; service managers and commissioners.

A purposive sampling approach (Palys, 2008) was adopted with an embedded chain-referral method used (Faugier and Sargeant, 1997). Eight CYP participants, and their families, from [REDACTED] ([REDACTED]) were invited via postal mail to attend the consultation event. Other stakeholders, such as professionals, were invited to attend via local and regional email distribution lists. All invitees were requested to disseminate the invitation to other potentially interested parties. To provide opportunity to capture as many people as possible, the consultation event was conducted over a five hour period, at a weekend, in a venue at a University that was accessible via personal and public transport.

Data collection tools and methods

The stakeholders came from diverse backgrounds in terms of age, ability, developmental level and professional background. Our approach needed to be engaging and pragmatic to allow for the sharing and evaluation of experiences as well as the identification of service and research priorities. Creative and art-based approaches are useful as sensitive and robust methods of collecting feedback, especially with CYP as thoughts and experiences that may prove too difficult to put into words can be expressed (Coad, 2007, Coad, 2012). Feedback from experiences of being in receipt, or delivering care, as well as priorities for future research and services were gathered using draw/write and tell techniques (Bradding and Horstman, 1999, Bagnoli, 2009) and group discussions (Krueger and Casey, 2014).

Procedure

The consultation exercise was facilitated by members of ‘the SCETCH project’ team ([REDACTED], [REDACTED] and [REDACTED]). A young person (aged 13) who had not experienced critical illness also helped

to facilitate the event to enhance the contribution of the younger CYP. The adult facilitators had the experience and skills required for working with and supporting a diverse group of stakeholders. The environment and structure of the event was organised to build an informal and supportive atmosphere as possible. The event was divided into two main activities:

(1) Introductions, aims and getting to know each other. The facilitators explained the purpose of the event which was delivered in a verbal and visual format. Collectively, ground rules were developed, agreed and documented to ensure all stakeholders were clear that they could share their experiences in a safe and confidential space (Krueger and Casey, 2014, Green and Thorogood, 2009). Attendees were encouraged to lead the discussions, ask questions and challenge others in a supportive and constructive manner.

(2) Evaluating experiences and identifying priorities. This activity took place in two separate rooms as follows: (a) Parents and health professionals with two facilitators (■■■ and ■■■); (b) CYP with two facilitators (■■■ and young person).

For each group questions were posed that included,

Question 1: "How can the needs of CYP and families be better supported/addressed?"

Question 2: "What should future projects/research focus on?"

Both groups culminated in a debrief exercise which included: (a) acknowledge anything they have learned from the event; (b) disclose anything that has not been covered during the other activities; (c) reflect on their experience of being involved in the project; (d) ask the facilitators any questions they may have.

Analysis

The data was collated, transcribed, and inputted into NVivo 11™. An inductive content analytical approach was used which is advocated when little is known about the phenomena or topic (Graneheim and Lundman, 2004, Elo and Kyngäs, 2008). The analysis process was conducted by two of the facilitators (██████████) with the data from each of the two groups separately (adults and then CYP). This involved: (1) immersing oneself in the data in order to comprehend the whole; (2) open coding and then grouping the codes into higher order headings called categories; and (3) comparing the groups through a conceptual map of the codes and categories (Vaismoradi et al., 2013, Elo and Kyngäs, 2008).

Ethical and research approvals

National Health Service (NHS) Research Ethics Committee (REC) permission was granted from East Midlands REC (ref: Derby 1, 12/EM/0230, UK) to invite CYP and families that had previously participated in ██████████ (██████████). However, as this was a consultation exercise in research, NHS research ethical approval was not required (National Institute for Health Research, 2014). Therefore stakeholders that were involved were not identified as research participants, but instead acted as ‘specialist advisors’. The term ‘advisor’ will be used in this paper.

Informed consent

Prior to any involvement in the consultation event, informed consent was obtained from all advisors. For those younger than 16 years, assent was obtained and informed consent from parent or legal guardian. It was explained that involvement was entirely voluntary and they could leave at any time. Throughout the consultation event the team strived to ensure that ethical standards were maintained including safeguarding advisors’ privacy. All team

members ([REDACTED] and [REDACTED]) had enhanced Disclosures and Barring Service (DBS) checks, and adhered to the Data Protection Act (1998).

Participant remuneration

In line with national guidance (INVOLVE, 2010), all advisors were provided a complimentary lunch and were remunerated for travel expenses.

Findings

In total 24 advisors attended the consultation event that included CYP (n=8 [including n=3 PICU survivors], aged 7-15 years); parents and carers (n=6); health professionals (n=8); commissioners and service managers (n=2). Findings from the analysis will be presented in relation to each of the questions posed to each group of advisors.

Question 2: How can the needs of CYP and families be better supported/addressed?

The adult group identified that CYP and families had a number of unmet needs following a childhood critical illness. It was therefore unanimous that there were many opportunities for CYP and families to be supported. Feedback identified a variety and range of support that was required during the short and longer-term following the survival of critical illness that included information, emotional, social and overall well-being. Adult advisors suggested that tools to catalogue, explore, and navigate recovery might be useful for both parents/carers as well as CYP, which could include a diary that mobilises art based methods. Some of the adults indicated that a care passport might also be useful for themselves as well as a communication aide between agencies and professionals. Feedback also included other forms of support and how they could be delivered. These included peer support, family

based, and professional delivered that could be delivered via a digital and/or an inter-personal/one-to-one platform.

Deficits identified by the adult group related to a lack of integration in care pathway that focused on individualised needs of the CYP survivor and their family. Feedback indicated that this could be addressed by better signposting and organisation of care and transitions. Suggestions included having a contact point with a professional who understood the child and that could be accessed irrespective of where the child or family were in the pathway. It was also indicated by the adult advisors that a case manager/facilitator might be useful in addressing this need.

For the CYP group feedback indicated that there was a clear focus on being “normal” and support needed to enable it. It was identified from the CYP data that their needs related to establishing their own position in world. Therefore the focus was on support to enable their own growth and gaining better understanding of situation to adapt and accept to where and who they are now. CYP suggested that family and friends were fundamental sources of support. However, they recognised that siblings currently did not have any support which needed to be addressed. Feedback from the CYP group also indicated that health care professionals, as well as online forums, could offer support and feedback to enable them to catalogue their journey the in long term.

It is evident from Figure 1 that by collating the categories and codes from the two groups convergence on how the needs of survivors and their families be supported. From this conceptual map it is apparent that both groups identified deficits in current provision. However the adult group focused on the resources and lack of integration in care pathway, whereas the CYP focused on support to enable being ‘normal’. That said, much overlap was

evident in the sub-categories/codes that identified that a mix of digital and interpersonal modes of delivering support would be acceptable and these could involve interfacing with a mixture of peer, family and professional for support.

Question 2: What should future projects/research focus on?

Feedback from the adult group identified that research needed to understand and explore, in order to support, the significant junctions in the CYP and families' journey following PICU. For adult advisors, developing interventions to facilitate and support transitions post-PICU was deemed important. These were specified as transitions from the PICU to ward to home and involved developing and testing of interventions delivered by health professionals to better support families. It was conveyed through the discussions that the focus of this support needed to be holistic and integrate the physical, psychological, and social domains.

Furthermore, respondents indicated that research needed to focus on enabling effective communication mechanisms throughout pathway/journey. Although the focus of the adults' feedback related to supporting the transitions in their care, it was also evident that there was a desire for future research to addressing unmet needs of CYP and families in the longer term post critical illness. It was recognised that this needed to be equitable for all CYP even if they appeared physically well.

For the CYP group feedback indicated that research should focus on developing surveillance and interventions to enable them to achieve their 'new normal'. CYP indicated that this should include more research on role of periodic follow up by health professionals and what impact providing feedback to CYP on how they are doing has on them. Furthermore, CYP suggested research needed to focus on supporting physical rehabilitation as well as developing ways in which CYP can access and engage with peer support.

It is evident from Figure 2 that compares codes and categories from the analysis of both groups that differing priorities as to the focus of future research are evident. For the adults it is evident that research should be focused on the processes of recovery, with particular interest in interventions to support the multiple transitions that may be faced by CYP survivors and their families. Interventions should be holistic in order to support the emotional as well as physical health and wellbeing of the CYP and family. Whereas the CYP group focused on the longer term outcome and interventions to maximise functional as well as social/emotional well-being.

Discussion

This is the first known published report of a consultation exercise that illuminates CYP and their families' voices as the foci for setting research priorities for PICU survivors.

Furthermore, as identified by Menzies et al. (2016) CYP PICU survivor and their sibling's views are extremely scant within the published literature. Therefore, this paper makes a contribution to liberating the perspectives of these stakeholders, and in turn offers a platform to help inform future service and research agendas.

The findings from this consultation exercise, indicate that future services, interventions and research must be holistic and family centred. Parents were keen that support services are child-centred, with CYP reciprocally identifying that family support was important and that siblings had unmet needs.

Findings identify that parents had unmet communication and information needs throughout their pathway and that future research as well as services should focus on addressing these. In particular, there remains a need for improved communication within PICU, centred on improving health care professionals and parent communication mechanisms (Meyer et al.,

2009). Moreover, communication approaches that embrace opportunities to better prepare and support families, on the ward, transitioning to home, and in the long term need exploration and further understanding.

A cross cutting tension exists between some CYP and families being empowered to self-manage, with others wanting services to be organized for them. This echoes individualized and empowerment agendas and we should be cognisant that one size may not fit all. An examination of societal and National Health Service (NHS) structure and individual agency might prove fruitful in examining such tensions (Taylor and Bury, 2007). Individualized needs require assessment via a documented process; this may be achieved by development of a validated tool to enable appropriate support and signposting to be implemented.

Both CYP and adult advisors identified a number of potential interventions that could help support and enhance the lives of PICU survivors. Some of these provide possible sustainable and simple solutions that empower families to self-manage and navigate pathways of care, such as an online forums for peer support, that do not require significant monetary resources (Eysenbach et al., 2004, Griffiths et al., 2006). However, such initiatives require robust testing as they have not previously been applied with this post-PICU population.

Furthermore, as indicated by Taylor and Butt (2000) pertinent outcome measures need to be identified or developed in order to assess effectiveness.

It was notable that CYP focused on longer term outcomes; this could be explained by an assumed lack of awareness of the PICU and hospital experience by admitted CYP. However, this hypothesis is not supported by contemporary empirical data from this patient population (Manning J.C, 2015). Instead it could be argued that CYP are focused on current needs and less bothered by the intricacies of their illness experience and the subsequent transitions.

Strengths and limitations

The core strength of this stakeholder event is that it included views from CYP PICU survivors, their siblings and parents/carers. Its methods were both structured and unstructured, participatory and inclusive. A wide range of stakeholders attended the event; health care professionals, CYP, service managers and health care commissioners; this legitimized the ideas, needs and priorities identified by the participating PICU CYP and families. However we are mindful that the participants were a self-selected sample and as such generalizability might be limited; however this was a PPI exercise as opposed to a piece of empirical research; the authors will expand on PPI echoing the INVOLVE (2012) cycle of research to increase transferability as far as possible. Moreover, it was noticeable that the participants did not fully embrace all minority ethnic groups. The participants were self-selecting so the ethnicity of the participants was outside our control but this highlights the need to improve PPI with minority groups of CYP and their families in future research. A final limitation was the homogeneity of the CYPs illness; this was a sub sample from a purposeful sample within a wider in-depth qualitative study (██████████) and this might have affected the resulting homogeneity of illness within the stakeholder event sample. Future quantitative work or the use of theoretical sampling in qualitative work will enable more representative heterogeneity of future samples. However, the priorities set within this stakeholder event remain reflective of contemporary empirical evidence by the current authors (██████████, ██████████, ██████████).

Conclusions

This consultation exercise provides further evidence as to the value of meaningful PPI in the development of priorities for research and healthcare services to ensure they are appropriate,

relevant and acceptable. This exercise clearly illuminates the importance of CYP PICU survivors in PPI as in some cases the priorities for them differed to those of adults, who are recognised in the literature and clinical practice as their proxy and advocate.

Consultation findings detail a number of important areas for future paediatric critical care research that focus on supporting transitions as well as the outcomes of CYP PICU survivors and their families. A number of potential sustainable interventions have been identified by CYP and families to meet their diverse needs following survival of critical illness which require development and testing. Furthermore, there is also definite scope for the development of a screening tool that discriminates between CYP and families, who may require support following PICU, and directs input required. In addition to the PICU academic community, we hope that the reported priorities will be of use to clinical staff, service providers and commissioners and those who have an interest in the reform of PICU services.

What is known about the subject?

- More children and young people are surviving childhood critical illness than ever before
- There appears to be a shift from the paediatric clinical and research communities to better understand and develop services and interventions to maximise the outcomes of PICU survivors
- Childhood critical illness survivors and their sibling's views and perspectives on the priorities for future research and services remain absent from the literature.

What this paper contributes?

- Outlines a pragmatic approach to undertaking meaningful patient and public involvement with multiple stakeholders that include CYP that have survived a critical illness/injury and their siblings.
- Presents priorities that have been identified by CYP PICU survivors, their parents/carers, their siblings' and other stakeholders for future research and services to focus on and develop.

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