

Factors affecting uptake of postmortem examination in the prenatal, perinatal and paediatric setting

C Lewis,^{a,b} M Hill,^{a,b} OJ Arthurs,^{b,c} C Hutchinson,^{b,d} LS Chitty,^{a,b} NJ Sebire^{b,d}

^a North East Thames Regional Genetics Service, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK ^b UCL Institute of Child Health, Great Ormond Street Institute of Child Health, London, UK ^c Department of Radiology, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK ^d Department of Histopathology, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK

Correspondence: N Sebire, Department of Paediatric Pathology, Camelia Botnar Laboratories, Great Ormond Street Hospital, London WC1N 3JH, UK. Email neil.sebire@gosh.nhs.uk

Accepted 8 February 2017. Published Online 21 March 2017.

Background Postmortem examination is the single most useful investigation in providing information to parents about why their baby or child died. Despite this, uptake remains well below the recommended 75%.

Objective To address the question ‘what are the barriers and motivators to perinatal, prenatal and paediatric PM examination?’

Search strategy Key databases including Pubmed and CINAHL; Cochrane library, websites of relevant patient organisations, hand search of key journals, first and last authors and references.

Selection criteria Peer-reviewed qualitative, quantitative or mixed methods research examining factors affecting uptake or decline of perinatal or paediatric postmortem examination.

Data collection and analysis Narrative synthesis; findings were compared across studies to examine interrelations.

Main results Seven major themes describing barriers to postmortem uptake were identified: dislike of invasiveness, practicalities of the procedure, organ retention issues, protective parenting, communication and understanding, religion and culture and professional or organisational barriers. Six major themes related to factors which facilitated parental consent were

identified: desire for information, contributing to research, coping and well-being, respectful care, minimally invasive options, and policy and practice. There were a number of themes in the literature that reflected best practice.

Conclusion Findings highlight the need for better health professional education and the fact some concerns may be mitigated if less invasive methods of postmortem were routinely available. New consent packages and codes of practice may have a positive impact on perception of examination after death. The landscape is changing; further research is necessary to assess the impact on postmortem uptake rates.

Keywords Autopsy, consent, decision, fetal, paediatric, perinatal, postmortem, qualitative, systematic review.

Tweetable abstract Systematic review to explore the barriers and motivators to perinatal, prenatal and paediatric postmortem examination.

Linked article This article is commented on by LJ Wimmer et al, p. 182 in this issue. To view this mini commentary visit <https://doi.org/10.1111/1471-0528.14829>.

Please cite this paper as: Lewis C, Hill M, Arthurs OJ, Hutchinson C, Chitty LS, Sebire N. Factors affecting uptake of postmortem examination in the prenatal, perinatal and paediatric setting. BJOG 2018;125:172–181.

Introduction

In the United Kingdom (UK) around one in 80 pregnancies results in termination following diagnosis of a fetal abnormality, stillbirth or neonatal death, representing at least 8000 cases per annum, and there are over 500 unexplained infant and childhood deaths annually.^{1–3} In these situations, postmortem (PM) examination is often required

to determine cause of death, provide recurrence risk, establish implications for family members, and direct management of future pregnancies.⁴ Evidence suggests that PM examinations result in clinically significant findings in 22–76% of cases depending on type of loss and is the single most useful investigation in providing information to parents about why their baby or child died.^{5,6} Moreover, several studies have shown a significant discrepancy between

the apparent clinical cause of death and the PM findings, emphasising the potential value of information derived from examination after death.⁷ Despite this, uptake remains well below the recommended 75%,⁸ with UK national data demonstrating that only 44% of stillbirths, 38% of perinatal deaths and 25% of neonatal deaths are followed by a consented PM examination.⁹

Traditional PM procedures have changed very little, typically employing large bodily incisions to allow access to internal organs for further analysis. However new, less invasive, methods of investigation after death have recently been developed in an attempt to improve PM examination rates.¹⁰ One promising approach is the use of cross-sectional imaging techniques, in particular magnetic resonance imaging (MRI) which can also be used to guide further tissue-sampling techniques.^{11,12} This approach was recommended as a realistic alternative to current invasive PMs in adults by the Department of Health PM, Forensic and Disaster Imaging Group in 2012 with the acknowledgement that there are important religious, cultural and humanitarian benefits offered by non-invasive PMs.¹³ Identifying the reasons why people accept or decline PM examinations from both the parental and professional perspective is vital to understand whether these might be mitigated by introduction of less invasive methods and also to identify other interventions that might support increased uptake. Although a growing number of studies have investigated parental and professional determinants of PM rates, there is only one published in-depth review, published more than a decade ago (2004). However, it was not conducted systematically and relates to PM in the context of clinical trials.¹⁴

The aim of this systematic review is to address the question 'what are the barriers and motivators to perinatal, pre-natal and paediatric PM examination?'

Methods

We followed the method described by The Centre for Reviews and Dissemination¹⁵ and the PRISMA checklist¹⁶ to conduct this systematic review. We undertook a quality assessment of the studies before conducting a narrative synthesis¹⁷ of the results. The initial search was undertaken in December 2015 and repeated in August 2016 (no new papers were identified).

Eligibility criteria

Included studies:

- Bereaved parents with experience of termination of pregnancy for fetal abnormality, stillbirth, neonatal or childhood death (<16 years), or health professionals or general public.
- Those where a diagnosis was known, e.g. childhood cancer, as well as where there was no confirmed diagnosis, in

order to explore whether motivations and barriers were similar or significantly different.

- Factors affecting uptake or decline of perinatal/paediatric PM examination.
- Qualitative, quantitative or mixed methods; in English and peer-reviewed.

Excluded studies:

- Included adult PM examination; uptake rates (unless they subdivide participant characteristics influencing PM rates); focus on verbal, social or psychological PM; bereavement studies.
- Non-English papers due resource constraints.
- Editorials, letters, abstracts or commentaries, non-research articles or case reports.

Search methods

To avoid publication bias, the search (conducted by C.L.) incorporated a variety of sources and methods. The search included:

- Electronic database search using CINAHL, PsychArticles, PsycINFO, PubMed and Web of Science;
- Cochrane Library, Google Scholar and websites of relevant patient organisations;
- Hand search of the first and last authors of the initial set of papers and reference lists of those papers;
- A hand search of the previous 5 years of publication of eight relevant journals;
- No time limit was set;
- Date related to autopsy/postmortem examination as defined by the studies; data were not available on specific components of the postmortem examination (such as placental examination, imaging) for the purposes of this study.

Search strategy

Search question

The SPIDER acronym is an established model for aiding systematic searches which include qualitative and mixed methods research.¹⁸ SPIDER was used to delineate the elements of the research question and search strategy (Appendix S1).

Study selection

Initial searches identified 1484 potential articles. Independent assessment (C.L. and M.H.) reduced this to 35 papers for quality assessment (Figure 1).

Quality assessment

The Centre for Reviews and Dissemination underscores the importance of assessing the quality of the research included in systematic reviews.¹⁵ We used the quality assessment tool described by Kmet et al.¹⁹ which provides

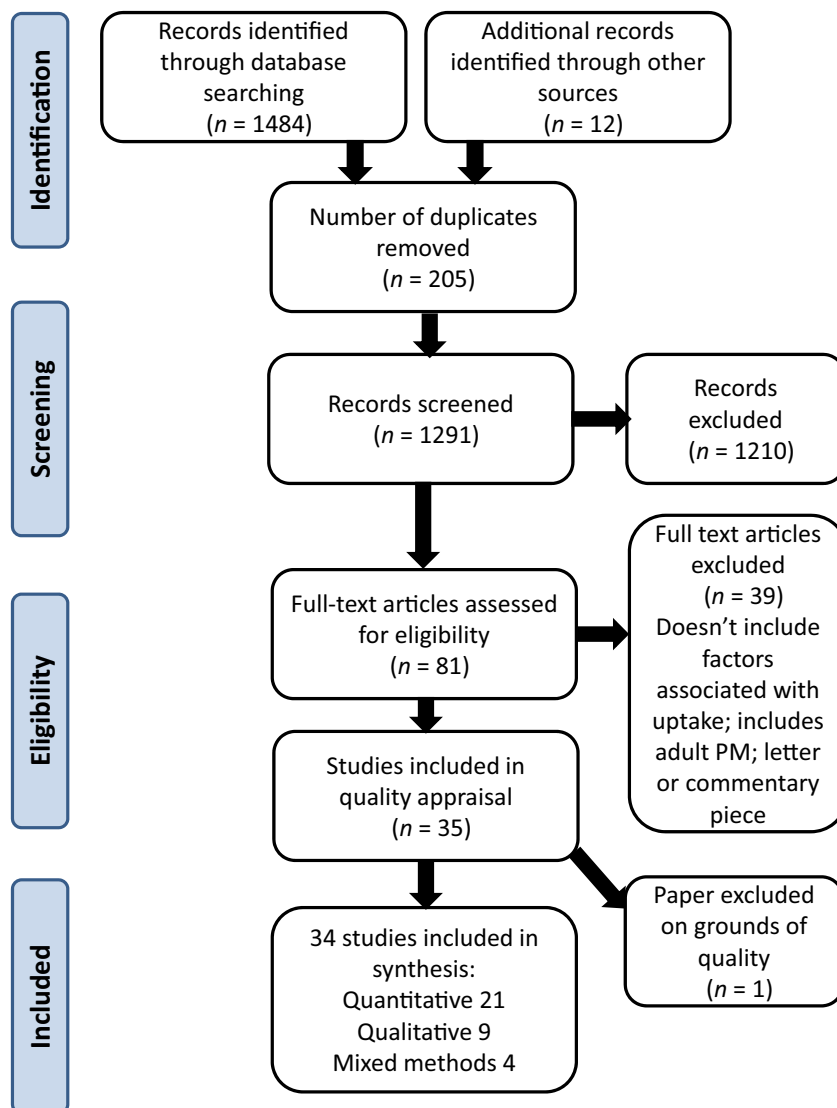


Figure 1. PRISMA 2009 flow diagram.

two sets of questions, one for qualitative and one for quantitative studies, the responses to which are converted into a percentage. A cut-off of 55%, described by Kmet et al.¹⁹ as liberal, was used. Using this methodology, one paper was excluded.

Data extraction

Key features of each study were extracted and tabulated (Table S1).

Synthesis

As both quantitative and qualitative studies with diverse approaches were included, a narrative synthesis was considered most appropriate. Using NVIVO 10 software, study findings were coded using the framework of barriers or

facilitators to PM uptake. For quantitative studies, statistical results and descriptions were tabulated and coded in terms of types of barriers or facilitators. For qualitative studies, direct quotes and descriptions were coded for all knowledge, beliefs, attitudes and practices in relation to the barriers and facilitators for PM. Findings were then compared across studies to examine interrelations, i.e. whether the same findings, concepts and theories existed across different studies, based on Noblit and Hare's method of meta-ethnography.²⁰ During this process it became apparent that a number of studies highlighted examples of what might be considered best practice, irrespective of whether people consented to PM. Therefore, a third overarching theme of 'best practice' was included to document these findings.

Results

Study and participant characteristics

An overview of the studies is presented in Table S1. Thirty-four studies published between 1982 and 2015 were included in the final review. The majority ($n = 26$) had been published since 2000. Twenty-one studies were quantitative: 12 were surveys^{21–32} and 9 were retrospective audits^{33–41}. Nine were qualitative consisting of semi-structured interviews,^{42–47} qualitative analysis of free-text in surveys,^{6,48} and focus groups.⁴⁹ Four were mixed methods.^{50–53}

Impact of parent or patient characteristics and experience on pm uptake

Twelve studies addressed the impact of parent or patient characteristics and PM uptake.^{23,26,33,36,38,40,41,43–45,51,53}

Three of four prenatal studies describing gestation showed that later gestation was significantly associated with increased PM uptake ($P < 0.001$, $P = 0.006$, $P = 0.016$)^{33,38,40} with the fourth indicating a significant association with earlier gestation ($P < 0.001$).²³ However, as the authors of this last study point out, one explanation for this finding may be the significant number of Muslim women (23%) in the study, for whom PM is permitted up to 120 days' gestation. In postnatal studies, consent was significantly more likely to be given for PM when babies or children were older in two USA studies ($P < 0.001$ and $P = 0.007$)^{36,40} but less likely in studies conducted in Zambia⁴³ and Scotland.⁴⁴ Known cause of death was also identified as a factor affecting PM uptake in three studies.^{40,44,51} Regarding parents' characteristics, ethnicity or parental educational level were not significant factors,^{23,36,38,53} but religion was positively related to the mother being non-Muslim in two studies ($P = 0.019$; $P = 0.007$).^{23,26}

Impact of health professional characteristics and experience on PM uptake

Six studies addressed the impact of health professional characteristics and experiences on PM uptake.^{21,23,27,31,39,53} Health professional role and experience were significantly associated with seeking consent,²⁷ perceived importance of PM⁵³, parental satisfaction with the consent discussion³¹ or PM uptake.^{23,39} Neonatologists and obstetricians were more likely to seek consent for PM than were neonatal nurses or midwives ($P < 0.001$), and neonatal nurses with more than 10 years' experience were more inclined to suggest PM than those with less experience ($P < 0.01$) in an Australian postal survey study.²⁷ Seniority was also a positive factor in a retrospective cohort study conducted in Scotland, in a Malaysian study where specialists were judged to have provided a 'good' explanation compared with house or medical officers,³¹ and in the USA, where advancement in staff position was associated with positive attitude regarding the importance of neonatal PM.⁵³

Conflictingly, in a UK study, Ben-Sasi et al.²¹ did not find any significant association between job type (physician versus nursing versus other) and PM acceptability. However, they did report that there were significant effects of demographic variables on the importance of factors which may prevent parental agreement for PM, including concern regarding possible disfigurement among non-white groups and concerns regarding delays to the funeral particularly for those of Indian, Asian or Arab ethnicity compared with white participants.

Barriers to uptake of PM

We identified seven major themes describing barriers to PM uptake: dislike of invasiveness, practicalities of the procedure, organ retention issues, protective parenting, communication and understanding, religion and culture, and professional or organisational barriers.

Dislike of invasiveness

A recurring theme related to parents' concerns about the invasiveness of the procedure^{25,29,45} and the baby's appearance following PM,^{21,29,44,46,49} as illustrated by one mother who declined PM because she would rather not know the cause of her baby's death than have her 'all cut up'.⁴⁵ Comparable results were found in quantitative studies, with concern about the examination of the baby a frequently cited reason for declining PM.^{25,44} Concerns around disfigurement, particularly among non-white responders, also existed in health professionals.²¹ Specifically, discussion around the removal of the brain was found to be distressing for parents⁴⁴ and professionals.⁴⁷

Practicalities of the procedure

Other factors included need to transfer babies to another hospital for PM, considered by 33% of parents but not health professionals as a strong barrier,²⁴ turnaround time for results,^{6,24} and the prohibitive cost or lack of insurance cover in the only international study where most respondents were from the USA.³²

Organ retention issues

Organ retention issues were described as a barrier to PM in six studies,^{6,21,24,34,39,46} with PM rates falling significantly for stillbirths and neonatal deaths since organ retention controversies (between 1996–2001) both in Australia³⁴ and in the UK,³⁹ following disclosures of unlawful organ retention. These findings are supported by a qualitative study in which a midwife commented that 'after the Alder Hey⁵⁴ episode many parents had been put off [PM] as they imagine parts of their baby sitting in specimen jars'.⁶ Negative press coverage was felt by 41–62% of health professionals to be a significant barrier to uptake in a study conducted in 2012,²⁴ although the majority (76%) of parents indicated that this factor had little influence on their decision.

Protective parenting

One of the main psychological barriers to uptake of PM was parents wanting to 'protect' their baby or child from unnecessary harm.^{43,45,46,49,51} Parents commented that their children had 'suffered enough',⁴³ were 'fragile like dolls',⁴⁶ and should be 'left in peace',⁵¹ with 'I already felt my baby had suffered enough' being the most frequently chosen response (44%) for respondents declining PM in a quantitative UK study.²⁹ Adopting this protective role has been suggested as one way parents could retain some degree of control over their situation.⁴⁵ Emotional distress during the PM discussion was also identified as a barrier.^{24,31,45,49} One parent described being an 'emotional wreck' and that being asked about PM was 'just too much all at once',⁴⁹ highlighting that the timing of the PM discussion is important. In a study on stillbirth, several women spoke of their fear that a PM would show that they were somehow to blame for their baby's death, although this theme did not occur elsewhere in the literature.⁴⁹

Communication and understanding

One of the most commonly cited barriers for both parents and health professionals was ambivalence about the value of the procedure.^{24,25,27,29,32,43,44,46,51} Twenty-six percent of PM decliners in a UK study²⁹ and 43% of decliners in a Zambian study⁴³ cited this as their reason for declining PM. Many midwives (35%) and obstetricians (32%) underestimated the value of PM in a UK study which reported a likelihood of <20% that useful information would be obtained.²⁴ Declining a PM because parents felt the cause of death was already known^{25,27,45,46,51} was cited in five studies, including those focused on stillbirth^{25,45} as well as childhood cancer.⁵¹ In one study on stillbirth, the authors note that for some parents a cause which was inferred during or just after birth was sufficient and accepted as true.⁴⁵

Embarrassment or feeling uncomfortable asking were also identified as barriers,^{6,24,42,47} with 'professional discomfort and lack of knowledge about the role and value of PMs' quoted in a qualitative study exploring attitudes of neonatologists and pathologists.⁴⁷ Lack of knowledge about the procedure, staff workload and lack of rapport with patients particularly among midwives, were also all identified as professional barriers to consenting parents in an internet-based survey about stillbirth.²⁴ Poor communication about the procedure was cited by both parents³¹ and health professionals³⁵. The quality and length of the consent form was cited by health professionals in two studies as a barrier to consent,^{6,47} particularly since the introduction of the NHS consent form, which it was felt was too long and drawn out.⁶ Timing of the information provision about PM was thought to be inappropriate by some women undergoing termination of pregnancy in an online survey.⁴⁸ Lack of trust of health professionals, either as a

consequence of the organ retention scandal or because parents thought a diagnosis should have been made prior to the child's death was also identified.^{6,43}

Religion and culture

Religious and cultural issues, including concerns about funeral delays and the cutting of the body being prohibited in Islamic law, were frequently discussed.^{6,22,24,25,29,31,43,45,46} A number of quantitative studies conducted in the UK, Ireland, Sweden and Zambia found that religion was not a frequently chosen reason for declining PM.^{22,25,29,43,45} However, in a study conducted in Malaysia,³¹ 28% cited this reason, possibly owing to the large Muslim population. In the study conducted in Zambia, nearly 10% of participants declined PM because of concerns that the mutilation of dead bodies would result in ancestral spirits making all women in their family infertile.⁴³

Professional or organisational barriers

A key barrier to PM concerned the lack of specialist training among healthcare professionals to consent patients, raised in five studies.^{6,21,24,42,47} Varying reports of levels of training reported among clinicians in two quantitative UK studies (21–82%)^{21,24} was supported by qualitative research conducted in the UK and USA.^{6,42,47} Similarly, Epstein commented that residents and fellows desired more guidance on the PM consent procedure.⁴²

Facilitators of PM consent

We identified six major themes related to factors which facilitated parental consent to PM: desire for information, contributing to research, coping and well-being, respectful care, minimally invasive options, and policy and practice.

Desire for information

Desire for information was a commonly noted factor in why parents consented to PM, with this being the most frequently cited reason in a number of quantitative studies.^{24,25,29,55} Parents wanted an explanation for what had happened^{22,24,25,27,29,44–46,49,52} and to understand the impact on future pregnancies,^{22,25,27,29,44,45,49} the latter being rated the most important in the study conducted by Breeze et al.²² on perinatal PMs. This was also a prominent theme in qualitative research, as highlighted by Meaney et al.,⁴⁵ who commented that 'all parents searched for meaning and aimed to uncover a reason why such an event had happened'.

Contributing to research

Advancement of medical knowledge^{22,24,25,29,50,52} and other altruistic motivations^{22,24,44,46,50–52} were frequently cited, as illustrated by a parent who commented that 'our heart-break could be somebody else's gain'.⁴⁴ The desire to

contribute to research was the dominant motivating factor for those parents who consented to PM where a diagnosis was already known.^{50–52} In one US study of parental attitudes to autopsies in children with lethal brain tumours, parents most frequently mentioned their wish to advance medical knowledge when asked why they agreed to PM.⁵⁰ Similarly, in a study on childhood cancer, most parents wanted to know how PM could help other children (90%) and how autopsy could help the medical team learn more about their child's cancer (90%).⁵²

Coping and well-being

Consenting to PM was identified as a coping strategy in two studies.^{25,46} Parents talked about 'getting a positive from a negative' in a qualitative study on parental attitudes⁴⁶ and 23 of 54 participants in a Swedish study of mothers' attitudes towards perinatal PM after stillbirth cited 'to better cope with the loss' as their reason for agreeing to PM.²⁵ Information to help provide 'closure' was cited by 10%²⁹ and 15%⁵⁰ of participants. A further benefit identified in two studies related to ruling out self-blame.^{29,45} Meaney et al.⁴⁵ identified that fathers wanted to rule out the potential that it was their genetic material that was a contributing factor to the death, and mothers with an antepartum stillbirth wanted to rule out the possibility that they had been negligent in some way during pregnancy.

Respectful care

Procedural factors associated with consenting to PM, familiarity with the PM procedure,⁴⁹ reassurance that the procedure would be carried out with care and dignity,⁴⁴ and knowing that their child's body would be respected were all cited as motivating factors.⁵²

Minimally invasive options

In four studies, the availability of a less invasive method of PM was found to be a motivating factor.^{21,22,26,47} One study reported that 46% of parents who refused conventional PM hypothetically consented to a minimally invasive option.²⁶ Notably, fewer than half (42%) of Muslim participants consented to conventional PM compared with 65% for a minimally invasive procedure, suggesting less invasive methods would be more acceptable to that religious group. A study of health professional views found that 40% thought minimally invasive PM was more acceptable than traditional PM ($P < 0.001$).²¹

Policy and practice

Two studies identified departmental policies as facilitating parental consent.^{6,39} One noted that improvement of PM rates coincided with relocation of perinatal pathology services to the same site as the obstetric unit, thereby improving local availability of specialist perinatal pathologists to

perform more timely autopsies.³⁹ In addition, perinatal pathology involvement in multidisciplinary meetings, case discussions and teaching was also found to improve staff perception of the value of PM. Finally, unit guidelines were changed to stipulate that only senior staff should offer PM, and an increase in the uptake of PM rates had been evident since that particular policy change.

Models of best practice

There were a number of themes in the literature that reflected what was considered best practice regardless of whether people consented to PM. These are highlighted in Figure 2.

Discussion

Main findings

Conducting research into parental experience and attitudes towards PM examination has been recognised as being particularly challenging due to the sensitivity of the subject matter.^{56,57} Despite these difficulties, these data indicate that researchers are attempting to meet these challenges, as a relatively large number of studies exploring the reasons parents accept and decline PM were identified. This systematic review highlights that current low PM uptake rates are a consequence of numerous factors encompassing not only procedural, psychological and cultural barriers from the parental perspective but also a number of professional barriers, many of which were common across countries. This is not surprising as parents are likely to have the same concerns around the invasiveness of the procedure and appearance of the body irrespective of where they live, their culture or religious beliefs. This review also identified examples of best practice that can provide valuable evidence to health professionals and policy makers about how these sensitive services should be delivered.

Strengths and limitations

The strengths of this review are the comprehensive search strategy used to identify papers and the use of a quality appraisal tool. Regarding limitations, older papers are not always well indexed, particularly if they are qualitative studies, and this is likely to be the reason why a significant number were identified through hand searching of authors and reference lists. The quality appraisal identified variability in the methodological rigour of the included studies, including low response rates, lack of detail regarding data analysis (particularly in qualitative studies) omission of statistical significance testing or analysis of the influence of variables such as age, education and ethnicity. Qualitative studies were predominantly limited to simple thematic analysis with very little interpretive content. Some papers included professional perceptions of parental views; these

- Having education materials available with words and phrases chosen that provide maximum comfort to the family^{21,50}
- National, standard information sheets and consent forms⁶
- Ample time given for discussion and questions about the PM procedure²²
- Having a trusted health professional who is understanding and empathetic to the parents' situation^{6,49,50}
- Training and support for staff to improve their knowledge and ability to guide and support parents⁶
- Explicitly building support amid the core labour group to release staff to spend time with bereaved women⁶
- Home visits to discuss PM and flexible time-lines for decision-making^{6,49,50,53}
- If considered appropriate, the option of discussing PM prior to termination of pregnancy, stillbirth or neonatal death or on multiple occasions before decision-making⁴⁹

Figure 2. Summary of findings relating to best practice when discussing PM with bereaved parents.

may not be consistent with the views of parents themselves, as indicated by the diversity in patient and professional views, as reported in one study describing the impact of the organ retention publicity.²⁴

Interpretation (findings in light of other evidence)

The concept of a less-invasive approach to PM is relatively new¹⁰ and has thus far only been considered in four studies, all of which found it to be more acceptable compared to traditional PM for most participants, particularly those of Muslim faith who have low rates of acceptance for traditional PM.^{21,26} This supports the view that less-invasive methods may be more acceptable to those parents who currently decline PM. Professional concerns around the limitations of the technology were raised^{6,22} highlighting that both advantages and limitations need to be made clear when discussing the various options for PM with bereaved families, which will require evidence based data related to particular clinical circumstances. Whilst non-invasive imaging-only approaches may provide useful additional information in some circumstances such as underlying structural malformations, they are unlikely in isolation to be useful for identification of many pathologies, such as metabolic diseases or infections.⁵⁸ However, when augmented with minimally invasive ancillary investigations (needle biopsy, placental examination etc.) overall accuracy rates similar to that of conventional PM (>90%) can be reached in many circumstances.⁵⁹ Further work with key stakeholders, particularly parents and community leaders from those religious groups who traditionally decline PM, would be valuable to determine whether less-invasive

methods would be religiously and culturally permissible and acceptable as part of routine clinical care. In addition, as less invasive methods of PM become increasingly available, research to explore whether the psychological barriers identified in this review remain prominent factors would be useful. Finally, future studies reporting on postmortem yield should provide data regarding which specific aspects of the procedure contributed to the diagnosis or main findings, in order to allow appropriate counselling of parents considering more limited approaches.

Health professionals' reluctance to raise the topic of PM was identified as a major barrier to uptake. Unless the death is being referred to the coroner, it is recommended that all parents should be offered the opportunity to discuss having a PM examination so that they can make an informed choice.⁶⁰ For this reason, creating environments that support health professionals to do this is critical. A number of studies identified examples of good practice when consenting parents for PM, underscoring the importance of national guidelines on best practice in this area. In the UK, the Health Tissue Authority (HTA) have recently addressed this issue with the introduction of codes of practice for PM examination.⁶¹ Many of the examples of best practice identified in this review are echoed in this document. The quality and appropriateness of the consent form and the need for national, standard information sheets and consent forms was also raised by health professionals.^{6,62} In 2013, the Stillbirth and Neonatal Death Society (SANDS) launched the Sands post mortem consent package, which was developed to provide information and guidance about PM for health professionals seeking

consent.⁶³ Research to determine the impact of this new consent package as well as the HTA guidance would be valuable to identify whether it has a significant impact on PM consent rates.

A notable facilitator of PM uptake was parental desire to contribute to research as this created an opportunity for the child's life to have meaning and value. Health professionals may be reluctant to ask bereaved parents about research as they are a potentially vulnerable group with high levels of distress, but it is clear that research is in fact an opportunity valued by many families. Some research has specifically focused on including bereaved parents in research studies,^{56,64,65} with one study reporting that 73% of parents stated taking part in research about PM decision-making had helped them feel better about the decision and regarded such studies as valuable and important.⁵⁶ These findings support the findings from this review and underscore the importance of discussing the potential for PM to contribute to medical knowledge when health professionals discuss the value of the procedure with parents.

Conclusion

In summary, this review provides an insight into the parent and professional barriers around consent to PM that have resulted in sub-optimal uptake rates. We have identified a number of important barriers including system level barriers within the NHS as well as practical, psychological and religious parental barriers that impact consent uptake. We have also identified a number of facilitators which highlight the need for better health professional education and the fact some concerns might be mitigated if less invasive methods of PM were routinely available to bereaved parents. Furthermore, new consent packages and HTA guidance may have a positive impact on perception of examination after death in the UK. The landscape is changing; further research is necessary to assess the impact on PM uptake rates.

Acknowledgements

This study was funded by an NIHR HTA award (14/168/02). OJA is an NIHR Clinician Scientist, and NJS and LC are NIHR Senior Investigators. NJS and JCH are supported by Great Ormond Street Hospital Children's Charity. This work was undertaken at Great Ormond Street Hospital, which received a proportion of funding from the United Kingdom Department of Health's NIHR Biomedical Research Centre funding scheme. This article presents independent research funded by the National Institute for Health Research (NIHR). The views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health.

Contribution to authorship

CL, LC and NS conceived of the review. CL and MH conducted the literature search and interpretation of data. Data interpretation was then discussed with LC, OA, CH and NS. CL drafted the first version of the review. MH, OA, CH, LC and NS contributed to revising it and producing the final draft and have approved the final version for publication.

Funding

This study was funded by an NIHR HTA award (14/168/02) 'Minimally invasive autopsy for fetuses and children based on a combination of post-mortem MRI and endoscopic autopsy examination.'

Details of ethics approval

This study received ethical approval from London Bloomsbury Research Ethics Committee on 4 April 16.

Disclosure of interests

None declared. Completed disclosure of interests form available to view online as supporting Information.

Supporting Information

Additional Supporting Information may be found in the online version of this article:

Table S1. Summary of Papers.

Appendix S1. Search strategy. ■

References

- 1 Office for National Statistics. *Childhood, Infant and Perinatal Mortality in England and Wales, 2012*. London: Office for National Statistics, 2015.
- 2 Office for National Statistics. *Unexplained Deaths in Infancy, England and Wales: 2013*. London: Office for National Statistics, 2015.
- 3 Royal College of Obstetricians and Gynaecologists. *Termination of Pregnancy for Fetal Abnormality in England, Scotland and Wales: Report of a Working Party*. London: Royal College of Obstetricians and Gynaecologists, 2010.
- 4 Lawn JE, Cousens S, Zupan J. 4 million neonatal deaths: when? Where? Why? *Lancet* (London, England) 2005;365:891–900.
- 5 Gordijn SJ, Erwich JJ, Khong TY. Value of the perinatal autopsy: critique. *Pediatr Dev Pathol* 2002;5:480–8.
- 6 Downe S, Kingdon C, Kennedy R, Norwell H, McLaughlin MJ, Heazell AE. Post-mortem examination after stillbirth: views of UK-based practitioners. *Eur J Obstet Gynecol Reprod Biol* 2012;162:33–7.
- 7 Shojania KG, Burton EC, McDonald KM, Goldman L. Changes in rates of autopsy-detected diagnostic errors over time: a systematic review. *JAMA* 2013;289:2849–56.
- 8 Royal College of Obstetricians and Gynaecologists and Royal College of Pathologists. *Fetal and Perinatal Pathology: Report of a Joint Working Party*. London: RCOG Press, 2001.

- 9 CEMACH. *Confidential Enquiry into Maternal and Child Health [CEMACH]. Perinatal Mortality 2007*. London: CEMACH, 2009.
- 10 Sebire NJ. Towards the minimally invasive autopsy? *Ultrasound Obstet Gynecol* 2006;28:865–7.
- 11 Breeze AC, Jessop FA, Set PA, Whitehead AL, Cross JJ, Lomas DJ, et al. Minimally-invasive fetal autopsy using magnetic resonance imaging and percutaneous organ biopsies: clinical value and comparison to conventional autopsy. *Ultrasound Obstet Gynecol* 2011;37:317–23.
- 12 Sebire NJ, Weber MA, Thayyil S, Mushtaq I, Taylor A, Chitty LS. Minimally invasive perinatal autopsies using magnetic resonance imaging and endoscopic postmortem examination ('keyhole autopsy'): feasibility and initial experience. *J Maternal-fetal Neonatal Med* 2012;25:513–8.
- 13 NHS Implementation Sub-Group of the Department of Health Post Mortem FaDIGP. *Can Cross-Sectional Imaging as an Adjunct and/or Alternative to the Invasive Autopsy be Implemented Within the NHS?* London: NHS, 2012.
- 14 Snowdon C, Elbourne DR, Garcia J. Perinatal pathology in the context of a clinical trial: a review of the literature. *Arch Dis Childhood Fetal Neonatal Ed* 2004;89:F200–3.
- 15 Systematic reviews. University of York; Centre for Reviews and Dissemination; 2009.
- 16 Liberati A, Altman DG, Tetzlaff J, Mulrow C, Gotzsche PC, Ioannidis JPA, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: explanation and elaboration. *BMJ* 2009;339:b2700.
- 17 Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006;3:77–101.
- 18 Cooke A, Smith D, Booth A. Beyond PICO: the SPIDER tool for qualitative evidence synthesis. *Qual Health Res* 2012;22:1435–43.
- 19 Kmet LMLR, Cook LS. *Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a Variety of Fields*. Alberta: Alberta Heritage Foundation for Medical Research, 2004.
- 20 Noblit GW, Hare RD. *Meta-Ethnography: Synthesizing Qualitative Studies*. London: Sage; 1988.
- 21 Ben-Sasi K, Chitty LS, Franck LS, Thayyil S, Judge-Kronis L, Taylor AM, et al. Acceptability of a minimally invasive perinatal/paediatric autopsy: healthcare professionals' views and implications for practice. *Prenatal Diagn* 2013;33:307–12 6p.
- 22 Breeze AC, Statham H, Hackett GA, Jessop FA, Lees CC. Perinatal postmortems: what is important to parents and how do they decide? *Birth* 2012;39:57–64.
- 23 Cannie M, Votino C, Moerman P, Vanheste R, Segers V, Van Berkel K, et al. Acceptance, reliability and confidence of diagnosis of fetal and neonatal virtuopsy compared with conventional autopsy: a prospective study. *Ultrasound Obstet Gynecol* 2012;39: 659–65.
- 24 Heazell AE, McLaughlin MJ, Schmidt EB, Cox P, Flenady V, Khong TY, et al. A difficult conversation? The views and experiences of parents and professionals on the consent process for perinatal postmortem after stillbirth. *BJOG* 2012;119:987–97.
- 25 Holste C, Pilo C, Pettersson K, Radestad I, Papadogiannakis N. Mothers' attitudes towards perinatal autopsy after stillbirth. *Acta Obstet Gynecol Scand* 2012;90:1287–90.
- 26 Kang X, Cos T, Guizani M, Cannie MM, Segers V, Jani JC. Parental acceptance of minimally invasive fetal and neonatal autopsy compared with conventional autopsy. *Prenatal Diagn* 2014;34: 1106–10.
- 27 Khong TY, Turnbull D, Staples A. Provider attitudes about gaining consent for perinatal autopsy. *Obstet Gynecol* 2011;97:994–8.
- 28 Landers S, Kirby R, Harvey B, Langston C. Characteristics of infants who undergo neonatal autopsy. *J Perinatol* 1994;14:204–7.
- 29 Rankin J, Wright C, Lind T. Cross sectional survey of parents' experience and views of the postmortem examination. *BMJ* 2002;324:816–8 3p.
- 30 Stolman CJ, Castello F, Yorio M, Mautone S. Attitudes of pediatricians and pediatric residents towards obtaining permission for autopsy. *Arch Pediatr Adolesc Med* 1994;148:843–7.
- 31 Valayatham V, Hiu J. Perinatal postmortem: factors influencing uptake and subsequent outcomes in an Asian population. *Med J Malaysia* 2012;67:87–90.
- 32 Warland J, O'Brien LM, Heazell AEP, Mitchell EA, Consortium S. An international internet survey of the experiences of 1,714 mothers with a late stillbirth: the STARS cohort study. *BMC Pregnancy Childbirth* 2015;15:172.
- 33 Brodlić M, Laing IA, Keeling JW, McKenzie KJ. Ten years of neonatal autopsies in tertiary referral centre: retrospective study. *BMJ (Clinical research ed)* 2002;324:761–3.
- 34 Khong TY, Tanner AR. Foetal and neonatal autopsy rates and use of tissue for research: the influence of 'organ retention' controversy and new consent process. *J Paediatr Child Health* 2006;42:366–9.
- 35 Khong TY, Mansor FA, Staples AJ. Are perinatal autopsy rates satisfactory? *Med J Aust* 1995;162:469–70.
- 36 Kumar P, Taxy J, Angst DB, Mangurten HH. Autopsies in children: are they still useful? *Arch Pediatr Adolesc Med* 1998;152:558–63.
- 37 Maniscalco WM, Clarke TA. Factors influencing neonatal autopsy rate. *Am J Dis Child (1960)* 1982;136:781–4.
- 38 Okah FA. The autopsy: experience of a regional neonatal intensive care unit. *Paediatr Perinat Epidemiol* 2002;16:350–4.
- 39 Stock SJ, Goldsmith L, Evans MJ, Laing IA. Interventions to improve rates of post-mortem examination after stillbirth. *Eur J Obstet Gynecol Reprod Biol* 2010;153:148–50.
- 40 Swinton CH, Weiner J, Okah FA. The neonatal autopsy: can it be revived? *Am J Perinatol* 2013;30:739–44.
- 41 Whitehouse SR, Kissoon N, Singh N, Warren D. The utility of autopsies in a pediatric emergency department. *Pediatr Emerg Care* 1994;10:72–5.
- 42 Epstein EG. End-of-life experiences of nurses and physicians in the newborn intensive care unit. End-of-life Experiences of Parents, Nurses & Physicians in the Newborn Intensive Care Unit. *J Perinatol* 2007;28:771.
- 43 Lishimpi K, Chintu C, Lucas S, Mudenda V, Kaluwaji J, Story A, et al. Necropsies in African children: consent dilemmas for parents and guardians. *Arch Dis Child* 2001;84:463–7.
- 44 McHaffie HE, Fowlie PW, Hume R, Laing IA, Lloyd DJ, Lyon AJ. Consent to autopsy for neonates. *Arch Dis Child* 2001;85:F4–7.
- 45 Meaney S, Gallagher S, Lutomski JE, O'Donoghue K. Parental decision making around perinatal autopsy: a qualitative investigation. *Health Expect* 2015;18:3160–71.
- 46 Snowdon C, Elbourne DR, Garcia J. Perinatal pathology in the context of a clinical trial: attitudes of bereaved parents. *Archives of Disease in Childhood – Fetal & Neonatal Edition* 2004;89:F208–11 1p.
- 47 Snowdon C, Elbourne DR, Garcia J. Perinatal pathology in the context of a clinical trial: attitudes of neonatologists and pathologists. *Archives of Disease in Childhood – Fetal & Neonatal Edition* 2004;89:F204–7 1p.
- 48 Fisher J, Lafarge C. Women's experience of care when undergoing termination of pregnancy for fetal anomaly in England. *J Reprod Infant Psychol* 2015;33:69–87.
- 49 Horey D, Flenady V, Conway L, McLeod E, Khong TY. Decision influences and aftermath: parents, stillbirth and autopsy. *Health Expect* 2014;17:534–44.

- 50 Baker JN, Windham JA, Hinds PS, Gattuso JS, Mandrell B, Gajjar P, et al. Bereaved parents' intentions and suggestions about research autopsies in children with lethal brain tumors. *J Pediatr* 2013; 163:581–6.
- 51 Sirkia K, Saarinen-Pihkala UM, Hovi L, Sariola H. Autopsy in children with cancer who die while in terminal care. *Med Pediatr Oncol* 1998;30:284–9.
- 52 Wiener L, Sweeney C, Baird K, Merchant MS, Warren KE, Corner GW, et al. What do parents want to know when considering autopsy for their child with cancer? *J Ped Hematol Oncol* 2014; 36:464–70.
- 53 VanMarter LJ, Taylor F, Epstein MF. Parental and physician-related determinants of consent for neonatal autopsy. *Am J Dis Child* 1987;141:149–53.
- 54 Batty D. Alder Hey report on use of children's organs 2001 [<https://www.theguardian.com/society/2001/jan/30/health.alderhey1>] Accessed 16 January 2017.
- 55 Wiener L, Sweeney C, Baird K, Merchant MS, Warren KE, Corner GW, et al. What do parents want to know when considering autopsy for their child with cancer? *J Pediatr Hematol Oncol* 2014;6:464–70.
- 56 Breeze AC, Statham H, Hackett GA, Jessop FA, Lees CC. Attitudes to perinatal postmortem: parental views about research participation. *J Med Ethics* 2011;37:364–7.
- 57 Dyregrov K. Bereaved parents' experience of research participation. *Soc Sci Med* 2004;58:391–400.
- 58 Addison S, Arthurs OJ, Thayyil S. Post-mortem MRI as an alternative to non-forensic autopsy in fetuses and children: from research into clinical practice. *Br J Radiol* 2014;87:20130621.
- 59 Thayyil S, Sebire NJ, Chitty LS, Wade A, Chong W, Olsen O, et al. Post-mortem MRI versus conventional autopsy in fetuses and children: a prospective validation study. *Lancet* (London, England) 2013;20:223–33.
- 60 Henley A, Schott J. Sands' learning outcomes for consent taker training: seeking consent/authorisation for a hospital post mortem examination of a baby. *J Neonatal Nurs* 2014;20:11–3.
- 61 Human Tissue Authority. Code of Practice 3: Post-mortem examination; 2016.
- 62 Snowdon C, Elbourne DR, Garcia J. Perinatal pathology in the context of a clinical trial: attitudes of neonatologists and pathologists. *Arch Dis Child Fetal Neonatal Ed* 2014;89:F204–7.
- 63 Sands: Stillbirth and Neonatal Death Society. Sands Post Mortem Consent Package; 2013.
- 64 Siassakos D, Storey C, Davey L. Stillbirth: public/patient involvement in sensitive research and research ethics. *BJOG* 2015;122:1111.
- 65 Snowdon C, Brocklehurst P, Tasker R, Plat MW, Harvey S, Elbourne D. Death, bereavement and randomised controlled trials (BRACELET): a methodological study of policy and practice in neonatal and paediatric intensive care trials. Southampton: NIHR Journals Library, 2014. July (Health Technology Assessment, No. 18.42).