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# Interventions for supporting parents' decisions about autopsy after stillbirth (Review)

Horey D, Flenady V, Heazell AEP, Khong TY

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[Intervention Review]

# Interventions for supporting parents' decisions about autopsy after stillbirth

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## ABSTRACT

### Background

Stillbirth remains one of the least understood areas of infant death and accurate data on the causes of stillbirth are the cornerstone of stillbirth prevention. An autopsy examination remains the gold standard post-mortem investigation for stillbirth. However, decisions about post-mortem investigations, particularly autopsy are difficult. The purpose of this review is to examine the effectiveness of methods to help parents who have experienced a stillbirth decide whether to have post-mortem investigations, including whether to have an autopsy performed.

### Objectives

The primary objectives were a) to examine the effectiveness of interventions to support parents' decisions about autopsy consent after a stillbirth on outcomes for parents, and b) to determine autopsy rates. Secondary objectives were to identify issues related to the acceptability of any interventions to parents and the feasibility of their implementation.

### Search methods

We searched the Cochrane Pregnancy and Childbirth Group's Trials Register (29 October 2012), the Cochrane Central Register of Controlled Trials (CENTRAL) (*The Cochrane Library* 2012, Issue 10), MEDLINE (1966 to 24 July 2012) and EMBASE (1980 to 24 July 2012), Current Controlled Trials metaRegister (mRCT) (18 September 2012) and the WHO International Clinical Trials Registry Platform Search Portal (ICTRP) (18 September 2012). We also searched the websites of the Stillbirth and Neonatal Death Charity (SANDS) and International Stillbirth Alliance (ISA) (18 September 2012) and then subsequently searched the websites of all the ISA member organisations.

### Selection criteria

Randomised controlled trials (RCTs) of interventions designed specifically to support parents who have experienced a stillbirth make decisions about their options for post-mortem investigations including all investigations after stillbirth compared with usual care.

### Data collection and analysis

Two review authors independently screened citations against the selection criteria.

**Main results**

No studies meeting the review inclusion criteria were identified. A search of 40 websites associated with supporting parents who experience stillbirth also found little reference to, or information about autopsy or other post-mortem examinations.

**Authors' conclusions**

Support for parents making decisions about autopsy or other post-mortem examinations after stillbirth must rely on the ad hoc knowledge and experience of those involved at the time.

## PLAIN LANGUAGE SUMMARY

**Interventions for supporting parents' decisions about autopsy after stillbirth**

Understanding the cause of a stillbirth is important to parents yet little is known how to help parents make difficult decisions about whether to have investigations carried out on their stillborn infant to help provide such information. These include autopsies, surgical investigations, imaging and other investigations. Information gained may help the bereaved parents to plan future pregnancies and assist in the management of these pregnancies. The findings would also add to research into the causes of stillbirth and the need to terminate pregnancies. Inadequate information and poor communication can lead some parents to avoid decisions or to regret making a decision not to have an autopsy examination.

The process for obtaining consent for such tests is difficult for both parents and health professionals so interventions that support decision-making are likely to be beneficial. Interventions of this type could ensure independent information is available to parents that may encourage discussion with health professionals and lead to greater involvement of parents in decision-making.

This review conducted an extensive search of the research literature but could find no randomised controlled studies that looked at interventions to support decision-making on autopsy or associated investigations. An additional search of 40 websites of parent and professional groups associated with an international stillbirth organisation found little information about autopsy or other relevant investigations. Parents who experience stillbirth need to know more about their options for investigating the cause of death. More research is needed to find how to support these difficult decisions in the best possible way.

## BACKGROUND

Stillbirth has enormous personal impact and is the most common cause of infant death (Cacciatore 2007; Cacciatore 2009) yet remains one of the least understood areas of childbirth. While other forms of infant death continue to decline in many regions, stillbirth rates remain virtually unchanged (Lawn 2011). Accurate data on the causes of stillbirth are the cornerstone of stillbirth prevention. An autopsy examination remains the gold standard post-mortem investigation for stillbirth (Flenady 2011a). In some countries autopsy is synonymous with post-mortem, although it more accurately applies to the surgical procedure undertaken by a pathologist to investigate causes of death. Autopsy and other post-mortem investigations can reveal the cause of a stillbirth or other important conditions, or rule them out, which can be important for informing future pregnancy care. However, decisions about post-mortem investigations, particularly autopsy are difficult, and are made by parents at one of the most difficult times in their lives (Oppewal 2001). Despite common perceptions, there is no actual prohibition to autopsy by any major religion (Davis 1996; Gordijin 2007). Similar reasons for poor consent rates to autopsy are found in both high-income and low-income countries: lack of resources (Lawn 2009; RCPAAWP 2004); lack of adequate information among family and health professionals (Khong 1997; Oluwasola 2009); and beliefs that no new information will be found (Cartlidge 1995; Lishimpi 2001). The purpose of this review is to examine the effectiveness of methods to help parents who have experienced a stillbirth to decide whether to have a post-mortem investigation, including whether to have an autopsy performed.

### Description of the condition

Understanding the cause of a stillbirth is important to most parents whose baby is stillborn. Explanation of stillbirth was described as a turning point in interviews with parents of stillborn infants, who felt medical investigations were needed to avoid any adverse impact on future pregnancies and to overcome the guilt felt by mothers (Säflund 2004). Similarly, fathers of stillborn babies found the "question of why" remained until "a definitive answer and an explanation of the cause" were given (Samuelsson 2001). Concern that raising such topics will add to parents' distress needs to be balanced with consideration of potential longer term positive effects (Brabin 1995) and the high importance parents place on knowing the cause of their infants' death (Gold 2007). The post-mortem investigation options available to parents to investigate stillbirth (Flenady 2011b), definitions of stillbirth (Lawn 2011) and legal requirements for consent for such investigations vary across countries, but in most countries decisions about autopsy following a stillbirth differ from those following neonatal death in one important way; the onus of the decision belongs to the parents alone. Parents may also need to decide whether to agree to other post-mortem investigations. These investigations can include maternal blood tests, amniocentesis following fetal death, baby-gram (full body X-Ray) or magnetic resonance imaging (MRI) of the baby (Flenady 2011b).

It can be difficult for health professionals to raise the issue of autopsy following a stillbirth (Khong 1997) and some are unprepared to care for parents when such events occur (Gold 2007; Hunt 2009).

## Description of the intervention

Interventions to support decision-making, such as decision aids, are intended to supplement advice provided by health professionals. Potential interventions are generally intended for choices considered values-sensitive or when the balance of benefits and harms are equivocal (Stacey 2009). In some cases consent forms could be designed to also deliver information, and may be considered a form of decision aid.

Interventions to support health decisions can be used independently, in conjunction with health professionals during clinical encounters or through mediated social encounters, such as a telephone decision coaching service (Elwyn 2010). They include decision aids, one-on-one counselling, group information or support sessions and decision protocols or algorithms designed for use in discussions with consumers. The aim of this review was to consider decision-support interventions for parents making a decision about an autopsy after a stillbirth.

An informational component is a necessary part of a decision-support intervention although there are no firm rules about what this should cover. It can include the context in which the decision is relevant, why a decision is required, the available options and their potential benefits and harms with the likelihood of such probabilities. The International Patient Decision Aids Standards (IPDAS) Collaboration states that decision aids should provide evidence-based information about a health condition, the options, associated benefits, harms, probabilities, and scientific uncertainties ( Elwyn 2006; Elwyn 2011; IPDAS 2005a; Stacey 2009 ).

Decision-support interventions commonly include what Elwyn 2010 termed deliberation components, which may be a values clarification exercise or guidance in decision-making.

## How the intervention might work

Current decision-making theories seek to describe the process of decision-making rather than explain how people might be supported to make health decisions (Elwyn 2011) and few decision-support interventions designed for consumers are explicitly based on decision-making theories or models (Bekker 1999; Durand 2008). However, as decision-support interventions focus on providing information, people who use them will be better prepared for decision-making, although information is not the only criterion people use when making health decisions (Bekker 2010; Elwyn 2009).

Interventions that can be used independently of health professionals may provide parents with types of information not otherwise available to them, whereas, those interventions designed for use in conjunction with others, such as clinicians, may encourage discussion and greater involvement in decision-making (Elwyn 2010).

The use of interventions to support decisions about autopsy after stillbirth may affect outcomes in different areas. Parents could be more certain that they have made the best decision for them, thereby reducing decisional conflict or uncertainty, and their needs for information may be better met. Decisions about any post-mortem investigations including autopsy may change, affecting autopsy rates and the types of post-mortem investigations performed. There is also likely to be an impact on costs, though this may be difficult to assess over the longer term.

## Why it is important to do this review

Stillbirths are an unaddressed global health problem and warrant thorough investigation (Bhutta 2011; Flenady 2011a; Frøen 2011; Goldenberg 2011; Lawn 2011; Pattinson 2011), which includes offering parents appropriate autopsy investigations within the resources of the setting in which the stillbirth occurs. For example, in high-income countries, the incidence of stillbirth in pregnancies that reach 22 weeks' gestation or more is one in 200, 10 times higher than sudden infant death (Frøen 2011; Smith 2007). Considerable variations occur in the uptake of perinatal autopsy even in resource-rich countries such as Australia, where autopsy rates after stillbirth in one state are more than double that of another. In Western Australia between 2005 and 2007 the majority of stillbirths (68.5%) were investigated with an autopsy, compared with only 30% in Queensland in 2009. Variation in uptake and quality

is important. Although not all post-mortem investigations can adequately explain the cause of a stillbirth, in a significant proportion of cases perinatal autopsies add additional information (Gordijin 2002; Michalski 2002), rule out possible causes and can even lead to changes of diagnosis (Wagner 2005). A comprehensive protocol for post-mortem investigations for stillbirth can reduce the lack of explanation to less than one in seven (Dickinson 2011; Flenady 2011a; Gordijin 2002).

Internationally, perinatal autopsies and other post-mortem investigations have been in decline over the past two decades (Brodlie 2002; Rose 2006) exacerbated by revelations of organ retention that occurred without explicit consent from families (Adappa 2007; Khong 2002; McHaffie 2001).

Reasons for decreasing autopsy rates across the world are likely to be complex and multi-faceted, and although research is limited, consent is considered a major factor (AHMAC 2002; Hull 2007; Khong 2006).

The process for obtaining consent for autopsy is difficult for both clinicians and parents. Decision-making is difficult for parents overwhelmed by grief and clinicians can feel inadequate talking to parents about the options for post-mortem investigation, sometimes because of their own misperceptions about perinatal autopsy. It would be helpful to both parents and health professionals to know how the process of decision-making can be effectively supported at such a difficult time.

The information gained from post-mortem investigations following stillbirth may assist in the planning and management of future pregnancies, add to research into the causes of stillbirth and may also reassure parents that they are not to blame and assist grief resolution (Flenady 2009). Inadequate information and poor communication can lead some parents to regret their decision about autopsy. Parents who do not have autopsy are more than twice as likely to regret their decision as those who elected to have the investigation (Rankin 2002).

It is important to review interventions that support decision-making for autopsy after stillbirth to identify optimal approaches based on the best available evidence.

# OBJECTIVES

To examine the effectiveness of interventions to support parents' decisions about autopsy consent after a stillbirth on outcomes for parents and to determine autopsy rates.

Secondary objectives are to identify issues related to the acceptability of any interventions to parents and the feasibility of their implementation.

# METHODS

## Criteria for considering studies for this review

### Types of studies

All published, unpublished, and ongoing randomised and quasi-randomised controlled trials with reported data.

### Types of participants

The primary participants will be parents who have experienced a stillbirth for births of 20 weeks' gestation or more. We will include health professionals as secondary participants.

### Types of interventions

Interventions will be designed specifically to support parents who have experienced a stillbirth make decisions about their options for post-mortem investigations including all investigations after stillbirth. We will compare interventions with usual care.

### Types of outcome measures

Primary and secondary outcomes will relate to parents. We will also consider the impact of the secondary outcomes for health professionals, including midwives, doctors and pathologists.

#### Primary outcomes

- Decisional conflict (using the Decisional Conflict Scale (O'Connor 1995))
- Information needs met (as defined by the study authors)
- Proportion and type of post-mortem investigations performed

#### Secondary outcomes

- Psychological outcomes (anxiety, depression, etc)
- Knowledge or understanding of options and possible outcomes
- Proportion and type of other post-mortem investigations performed
- Cost

## Search methods for identification of studies

### Electronic searches

We contacted the Trials Search Co-ordinator to search the Cochrane Pregnancy and Childbirth Group's Trials Register (29 October 2012).

The Cochrane Pregnancy and Childbirth Group's Trials Register is maintained by the Trials Search Co-ordinator and contains trials identified from:

1. monthly searches of the Cochrane Central Register of Controlled Trials (CENTRAL);
2. weekly searches of MEDLINE;
3. weekly searches of EMBASE;
4. handsearches of 30 journals and the proceedings of major conferences;
5. weekly current awareness alerts for a further 44 journals plus monthly BioMed Central email alerts.

Details of the search strategies for CENTRAL, MEDLINE and EMBASE, the list of handsearched journals and conference proceedings, and the list of journals reviewed via the current awareness service can be found in the 'Specialized Register' section within the editorial information about the Cochrane Pregnancy and Childbirth Group.

Trials identified through the searching activities described above are each assigned to a review topic (or topics). The Trials Search Co-ordinator searches the register for each review using the topic list rather than keywords.

In addition, we searched the CENTRAL (*The Cochrane Library* 2012, Issue 10), MEDLINE (1966 to 24 July 2012) and EMBASE (1980 to 24 July 2012) See Appendix 1 for search strategies used. We also searched for unpublished and ongoing studies in the following registry search platforms: Current Controlled Trials metaRegister (mRCT) and the WHO International Clinical Trials Registry Platform Search Portal (ICTRP) (18 September 2012).

### Searching other resources

If we had identified any studies for inclusion, we intended to conduct author and citation searches in Science Citation Index database and screen the reference lists of all included studies to identify other possible trials and for any concurrent qualitative studies. We also searched on-line resources available through consumer and other organisations, such as the Stillbirth and Neonatal Death Charity (SANDS) and the International Stillbirth Alliance (ISA) (18 September 2012). These included the websites of the member organisations of the (ISA). Post-hoc, we also included a search of these websites for any information about stillbirth and autopsy (see Table 1).

### Correspondence

We intended to write to the corresponding authors of any included studies and relevant reviews to assist with identification of unpublished and ongoing studies.

We did not apply any language restrictions.

## Data collection and analysis

We did not identify any trials for inclusion in this version of the review, but if trials are included in updates we will use the methods set out in Appendix 2.

# RESULTS

## Description of studies

### Results of the search

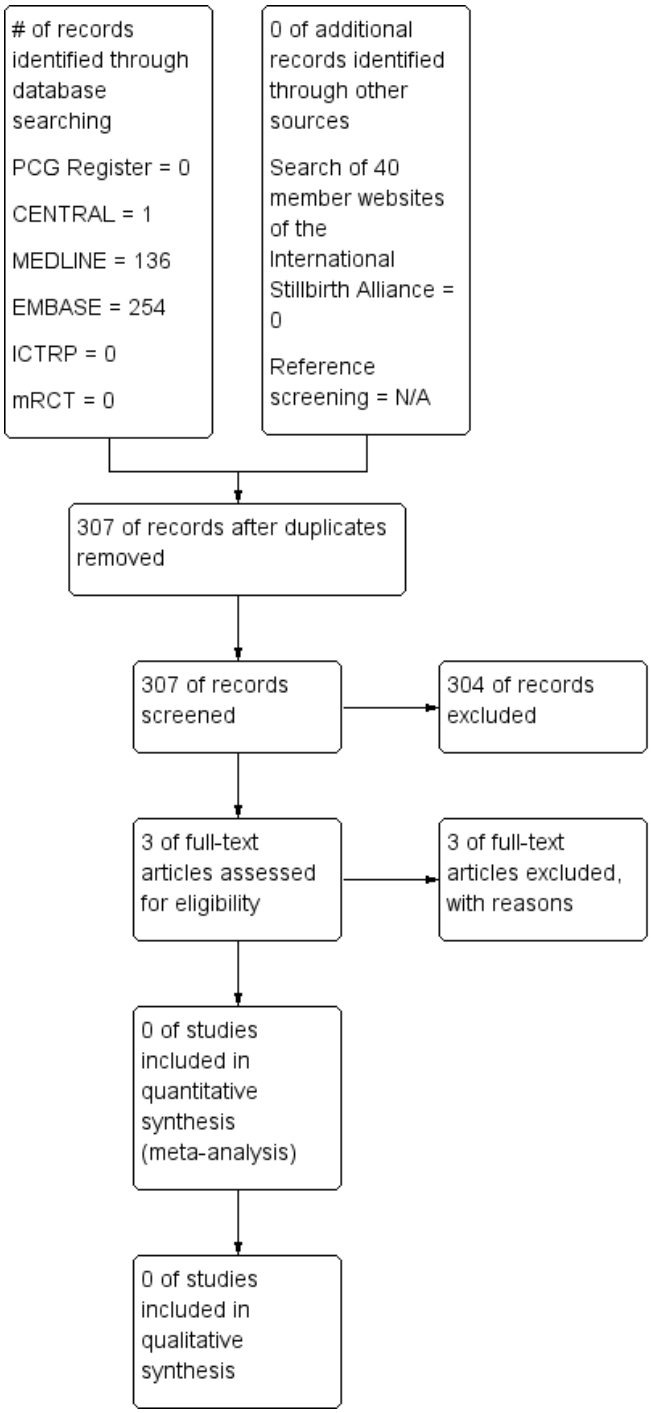
There were no relevant trial reports in the Cochrane Pregnancy and Childbirth Group's Trials Register. Additional searches of CENTRAL, MEDLINE and EMBASE identified 307 titles after the removal of duplicates. We retrieved three articles for full review. There were also no relevant trial reports from in a search of 40 websites of listed members of the International Stillbirth Alliance (see Table 1).

A post hoc search of these websites for reference to, or information about, autopsy or post-mortem examinations found little relevant information. Only 13 of the 40 websites included any reference to autopsy after stillbirth and only four websites provided any explanation of an autopsy (see Table 1). Two websites indicated that an autopsy after stillbirth was a parental right.

### Included studies

There were no included studies - see Figure 1.

**Figure 1. Study flow diagram.**



### Excluded studies

Three studies retrieved for full review were excluded (see Characteristics of excluded studies tables).

### Risk of bias in included studies

There were no included studies.

### Effects of interventions

There were no included studies.

## DISCUSSION

Parents who experience stillbirth face difficult decisions about post-mortem investigations but there are no studies that have examined the effectiveness of ways to support their decision-making. There also appears to be little information readily available to parents about this issue. Health professionals can provide information and support to parents about autopsy but some may lack adequate information (Khong 1997) or perceive barriers to counselling, such as lack of rapport, staff workload or religious or cultural reasons that may be less significant or important to parents. Conversely, health professionals may not recognise service-based factors, such as the lag time for results or the need to transfer babies to another centre for an autopsy that can be important to parents (Heazell 2012). The authors are aware that individual hospitals may have their own leaflets to give information to parents, but there is little evidence to guide the content of such information or assessment of its impact. However, it was impossible to identify, collect and review each individual hospitals information; standardised information for regions and/or nations would make this task easier for future reviews.

### Summary of main results

There were no included studies.

### Overall completeness and applicability of evidence

There were no included studies.

### Quality of the evidence

There were no included studies.

### Potential biases in the review process

Despite an extensive search we found no studies.

### Agreements and disagreements with other studies or reviews

A retrospective cohort study identified an association between policy changes in an obstetric unit and acceptance of post-mortem examinations (Stock 2010). These changes included easier access to perinatal pathology, new guidelines for management following stillbirth, and the introduction of an education program for medical and midwifery staff about the importance of autopsy. All parents who experience stillbirth are now offered post-mortem examination by consultants or senior registrars (more than five years postgraduate experience) with a prospective audit in place to monitor staff involvement and consent rates. In addition every stillbirth is reported at monthly perinatal mortality meetings and staff are encouraged to attend autopsies (Stock 2010).
A small study of 35 parents who had experienced perinatal death (including 16 from stillbirth) were asked about attitudes to, and expectations of, post-mortem examinations. The authors concluded that the desire for information about the cause of their baby's death outweighed possible barriers to consenting. A significant proportion of those surveyed (16/35) reported that completing the questionnaire made them feel better about their decision (Breeze 2012).
A recent UK survey of health professionals and parents who had experienced stillbirth provides further evidence of the need for support in decision-making around this issue. More than one in five parents responding to the survey were dissatisfied with their decision (21%, 95% confidence interval (CI) 17.3% to 24.7%), with a large majority of these wanting more investigations (90%, 95% CI 80.3% to 93.7%). Parents who did not have an autopsy were twice as likely to be dissatisfied with their decision than those who did (odds ratio 2.43, 95% CI 1.53 to 3.87) (Heazell 2012).These studies suggest that support with decision-making is likely to benefit parents.

## AUTHORS' CONCLUSIONS

## Implications for practice

As no studies were identified, there is no evidence about how to effectively support parents' decision-making about autopsy consent and post-mortem examinations after stillbirth.

## Implications for research

All aspects of support for parents' decision-making related to post-mortem examinations after stillbirth require further study. This includes strategies to address parent, health professional and institutional barriers to understanding the role of autopsy and practices associated with it. The challenges of conducting randomised controlled trials in this area suggest that meta-analyses will be necessary to resolve important questions. This could be facilitated by developing consensus on study parameters and appropriate outcome measures and their definitions.

# ACKNOWLEDGEMENTS

As part of the pre-publication editorial process, this review has been commented on by three peers (an editor and two referees who are external to the editorial team), a member of the Pregnancy and Childbirth Group's international panel of consumers and the Group's Statistical Adviser.

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* *Indicates the major publication for the study*

## CHARACTERISTICS OF STUDIES

### Characteristics of excluded studies *[ordered by study ID]*

| Study | Reason for exclusion |
|---|---|
| Breeze 2012 | Not a randomised controlled trial. |
| Rowland 2009 | Not a randomised controlled trial. |
| Stock 2010 | Not a randomised controlled trial. |

## DATA AND ANALYSES

This review has no analyses.

## ADDITIONAL TABLES

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country**

| Country | Name | Description | URL | Reference to or information about autopsy or postmortem examinations? |
|---|---|---|---|---|
| Argentina | Era en Abril | First non-profit organisation in Latin America that provides assistance to parents of babies who died during pregnancy, childbirth or after birth | http://www.eraenabril.org | Parental rights when a baby dies include being able to order an autopsy and for information provided in easy to understand terminology (including autopsy and pathology reports) |
| Australia | Australian and New Zealand Stillbirth Alliance (ANZSA) | Member-driven organisation focused on preventing stillbirth in Australia and New Zealand | http://www.stillbirthalliance.org.au | Explanation of autopsy included in "Resources for parents". |
| Australia | Australian College of Midwives (ACM) | National, not-for-profit organisation that serves as the peak professional body for midwives in Australia | http://www.midwives.org.au | None found. |
| Australia | Bears Of Hope | Offers support and guidance for parents who experience the loss of their baby during pregnancy, birth or infancy through the donation of a teddy bear | http://www.bearsofhope.org.au | Stillbirth information includes explanation of autopsy and the associated processes |
| Australia | Perinatal Society of Australia and New Zealand (PSANZ) | Multidisciplinary society dedicated to improving the health and long-term outcomes for mothers and their babies | http://www.psanz.com.au/ | None found. |
| Australia | Pregnancy Loss Australia | Bear-giving program and counselling services to support parents and families who have experienced any gestation loss of a baby | http://www.teddyloveclub.org.au/ | Mentioned in several parent stories of their pregnancy loss experience |

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country** (*Continued*)

| | | | | |
|---|---|---|---|---|
| Australia | Royal Australian and New Zealand College of Obstetricians and Gynaecologists (RANZCOG) | Medical College. | http://www.ranzcog.edu.au/ | Found in news reports. Media Wrap Up includes link to national radio program on topic of stillbirth autopsy |
| Australia | Royal Australian College of General Practitioners (RACGP) | Australia's largest professional general practice organisation and represents urban and rural general practitioners | http://www.racgp.org.au/ | Only in reference to SIDS. |
| Australia | SANDS Australia | National charity organisation of state-based parent-man-<br>aged, not-for-profit associations that aim to facilitate healthy grieving following the death of a baby through miscarriage, stillbirth, newborn death or termination | http://www.sands.org.au/ | FAQ include *"I had a stillbirth and the hospital did an autopsy - is that normal?* ". |
| Australia | SIDS and Kids Australia | National not-for-profit organisation with history of health promotion, bereavement support, advocacy and research | http://www.sidsandkids.org/ | Brief explanation of autopsy/post-mortem included in information about stillbirth and miscarriage<br>Organisation also hosted national pathology workshop about SIDS and autopsy |
| Australia | Stillbirth Foundation | Funds and encourages research into stillbirth and works to increase public awareness about stillbirth | http://www.stillbirth-foundation.org.au/ | Information about funded autopsy studies included under Research |
| Canada | ParentCare | Support group of parents who have suffered the loss of a baby through miscarriage, ectopic pregnancy, stillbirth or early infant death up to 28 days | http://www.parent-care.ca/ | None found. |
| Canada | Walk to remember | Annual event for families who have lost a baby by miscarriage, stillbirth, early infant death or SIDS | http://www.walktoremember.ca/ | None found. |

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country** (*Continued*)

| | | | | |
|---|---|---|---|---|
| | | to celebrate and honour these babies | | |
| Denmark | Landsforgeningen Spadbarnsfonden | Non-profit organisation that aims to support those who have lost children, and work to prevent the further infant deaths | http://www.spadbarnsfonden.se/ | None found. |
| International | International Society for the Study and Prevention of Perinatal and Infant Death (ISPID) | Not-for-profit organisation with a mission to advance research and increase knowledge in areas of perinatal and infant health and mortality. Also aims to serve as a centralised resource for sharing of information worldwide and connecting organisations and individuals | http://www.ispid.org/ | FAQ on stillbirth includes "*Are autopsies important? If so, why?".* |
| International | International Stillbirth Alliance (ISA) | Non-profit coalition of organisations dedicated to understanding the causes and prevention of stillbirth | http://www.stillbirthalliance.org/index.php | None found. |
| Ireland | A Little Lifetime Foundation | Voluntary organisation found by group of bereaved parents whose babies died before or at birth (stillbirth) or sometime after birth (neonatal death) | http://www.alittlelifetimefoundation.ie/ | None found. |
| Italy | Ciao Lapo Onlus | Scientific and welfare association comprised of physicians, psychologists, midwives and parents who have dealt with the experience of illness and loss in pregnancy or after birth that offers psychological and psychosocial support to parents and families who are experiencing or have experienced high-risk pregnancies, diagnosis of fetal pathology of their children, and the loss of a | http://www.ciaolapo.it/ | Assumed - advice includes *"any photo taken before the autopsy".* |

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country** (*Continued*)

| | | | | |
|---|---|---|---|---|
| | | child during pregnancy | | |
| Japan | Japan Academy of Midwifery (CAM) | To advance the science of midwifery to raise the standard of health care provided by professional midwives to mothers, babies and families, and women in every stage of life | http://square.umin.ac.jp/jam/english.html | None found. |
| Japan | Luke's Group for Parents of Angels | Support group for parents who have lost a child to neonatal death, stillbirth or abortion | http://plaza.umin.ac.jp/artemis/rcdnp/tenshi/index.html | None found. |
| Japan | SIDS Family Association Japan | Voluntary organisation to support parents who experience SIDS | http://www.sids.gr.jp/ | None found. |
| Japan | With Angels in the Sky | | http://www.h4.dion.ne.jp/~wais.kt/ | Unclear. |
| New Zealand | SIDS New Zealand | National organisation established to provide services for families and communities who have had children of any age die suddenly and/or unexpectedly of any cause including SIDS | http://www.sids.org.nz/ | None found. |
| Norway | Norwegian SIDS and Stillbirth Society (Landsforeningen uventet barnedød) | National association to support parents who have lost an infant unexpectedly including those who died in pregnancy | http://www.lub.no/ | None found. |
| Paraguay | Juan Pablito | Foundation to improve the care of babies with trisomy 13 and trisomy 18 and other premature babies and their families and to raise awareness about the death of babies during pregnancy, labour and after birth | http://www.juanpablito.com/ | Parental rights when a baby dies include being able to order an autopsy and for information provided in easy to understand terminology (including autopsy and pathology reports) |

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country** (*Continued*)

| | | | | |
|---|---|---|---|---|
| Spain | U MAMANITA | Parent organisation to support and inform mothers and fathers in Spain who experience infant death or stillbirth | http://www.umamanita.es | Under "Practical things but important" there is information about decisions and processes around autopsy |
| Sweden | Spädbarnsfonden | Non-profit organisation that aims to support those who have lost children, and work to prevent the further infant deaths | http://www.spadbarnsfonden.se/ | Unclear. |
| The Netherlands | The Fetal Medicine Foundation Netherlands | Organisation for health professionals that strives for standardisation of research and education during pregnancy by offering | http://www.fetalmedicine.nl/ | None found. |
| UK | National Perinatal Epidemiology Unit (NPEU) | Multidisciplinary research team dedicated to improving the care provided to women and their families during pregnancy, childbirth and the postpartum period, as well as the care provided to the newborn | https://www.npeu.ox.ac.uk/ | None found. |
| UK | Royal College of Obstetricians and Gyaecologists | Medical College. | http://www.rcog.org.uk/ | N (only related to maternal death). |
| UK | Sands UK | National charity established by bereaved parents that aims to support anyone affected by the death of a baby, work with health professionals to improve care and promote research and changes in practice that reduce loss | http://www.uk-sands.org/ | None found. |
| USA | 1$^{st}$ Breath | Provides education, advocacy, and public awareness of stillbirth in addition to assisting families and medical professionals dealing with the death of a baby | http://www.1stbreath.org | Includes articles that refer to autopsy. |

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country** (*Continued*)

| | | | | |
|---|---|---|---|---|
| USA | A Place To Remember | Publishes and provides uplifting support materials and resources for those who have been touched by a crisis in pregnancy or the death of a baby | http://www.aplacetoremember.com | None found. |
| USA | Angel Names Association | Nonprofit charitable organisation dedicated to assisting families of stillborn children through programs designed to provide financial assistance for end-of-life expenses and counselling services, and funding for stillbirth research | http://www.angelnames.org/ | None found. |
| USA | First Candle | National non-profit health organisation uniting parents, caregivers and researchers nationwide with government, business and community service groups to advance infant health and survival. | http://www.firstcandle.org/ | Included in parent story about SIDS. |
| USA | Global Alliance to Prevent Prematurity and Stillbirth (gapps) | Focus on achievement of the Global Action Agenda and United Nations Millennium Development Goals 4 and 5. Work to strengthen collaborations in maternal, newborn and child health, and develop a unified, global focus on preterm birth and stillbirth | http://gapps.org/ | None found. |
| USA | Hygeia 2012 | Hygeia 2012 is founded on principals derived from a personal philosophy to always maintain, respect and teach the tenets that the trust inherent in the doctor-patient relationship | http://drberman.org/abouthygeia2012.htm | None found. |

**Table 1. Website search of International Stillbirth Alliance (ISA) members by country** (*Continued*)

| | | | | |
|---|---|---|---|---|
| USA | Neo-Fight | Non-profit organisation dedicated to helping families experiencing a perinatal crisis which began as a parent support group | http://www.neofight.org/ | None found. |
| USA | Pre-Vent | Non-profit charitable organisation comprised of trained healthcare providers and administrators that advances the education of skilled birth attendants and community health workers and promotes preventive health measures among the vulnerable and poor populations | http://www.pre-vent.org | None found. |
| USA | Star Legacy Foundation | Non-profit organisation that raises funds to support stillbirth research and education | http://www.starlegacyfoundation.org | None found. |

SIDS: Sudden Infant Death Syndrome

# APPENDICES

## Appendix 1. Search strategies

CENTRAL (*The Cochrane Library* 2012, Issue 10) (Wiley interface)
#1 MeSH descriptor: [Stillbirth] this term only
#2 stillbirth or still-birth or stillborn or still-born or "still birth" or "still born"
#3 MeSH descriptor: [Fetal Death] explode all trees
#4 "fetal death" or "foetal death" or "fetal loss" or "foetal loss"
#5 MeSH descriptor: [Autopsy] explode all trees
#6 postmortem or post next mortem or post-mortem
#7 MeSH descriptor: [Decision Making] explode all trees
#8 MeSH descriptor: [Counseling] explode all trees
#9 MeSH descriptor: [Algorithms] explode all trees
#10 decision or inform* or guide or guidance or support or decide
#11 #1 or #2 or #3 or #4
#12 #5 or #6
#13 #7 or #8 or #9 or #10
#14 #11 and #12 and #13

MEDLINE (via OVID) (1966 to 24 July 2012)
1 Stillbirth/
2 exp Fetal Death/
3 (stillbirth or still-birth or stillborn or still-born or “still birth” or “still born”).ti,ab.
4 (“fetal death” or “foetal death” or “fetal loss” or “foetal loss”).ti,ab.
5 Autopsy/
6 (postmortem or post-mortem or “post mortem”).ti,ab.
7 exp Counseling/
8 exp Decision Making/
9 algorithms/
10 (decision* or inform* or guide or guidance or support* or decide).ti,ab.
11 1 or 2 or 3 or 4
12 5 or 6
13 7 or 8 or 9 or 10
14 11 and 12 and 13
EMBASE (via NHS Evidence)
1. STILLBIRTH/
2. (stillbirth OR still-birth OR stillborn OR still-born OR “still birth” OR “still born”).ti,ab
3. AUTOPSY/
4. CAUSE OF DEATH/
5. (postmortem OR post-mortem OR “post mortem”).ti,ab
6. exp COUNSELING/
7. ALGORITHM/
8. DECISION MAKING/
9. (decision* OR inform* OR guide OR guidance OR support* OR decide).ti,ab
10. (fetal ADJ loss OR foetal ADJ loss OR fetal ADJ death OR foetal ADJ death).ti,ab
11. 1 OR 2 OR 10
12. 3 OR 4 OR 5
13. 6 OR 7 OR 8 OR 9
14. 11 AND 12 AND 13
**WHO International Clinical Trials Registry Platform Search Portal** (ICTRP) **and Current Controlled Trials metaRegister** (mRCT) (18 September 2012).
stillbirth AND decision(s) AND autopsy
stillbirth AND counseling AND autopsy
stillbirth AND decision(s) AND postmortem
stillbirth AND counseling AND postmortem
fetal death AND decision(s) AND autopsy
fetal death AND counseling AND autopsy
fetal death AND decision(s) AND postmortem
fetal death AND counseling AND postmortem

## Appendix 2. Proposed data extraction and management

### Data extraction and management

#### Quantitative data

We will design a form to extract data. For eligible studies, two review authors will extract the data using the agreed form. We will resolve discrepancies through discussion or, if required, we will consult a third author. We will enter data into Review Manager software (Revman 2011) and check for accuracy.

The data extraction form will include the following components:

### Details of study

**Study design:** description of comparison group; description of usual care comparison; aim of study; methods of recruitment of participants; inclusion/exclusion criteria for participation in study; informed consent obtained (yes/no/unclear); ethical approval (yes/no/unclear); funding source and amount (if stated); statistical methods and their appropriateness (if relevant) and consumer involvement (in the design of study and/or intervention, in delivery of intervention, in evaluation of intervention, in interpretation of study findings).
*Intervention quality:* any information on the quality of the intervention as assessed by the study authors; including information related to the fidelity/integrity of the intervention, such as if it was delivered as intended or not, and rate of attrition.
For *methodological quality* of the study, we will use the seven domains of the 'Risk of bias' assessment tool (see 'Assessment of risk of bias in included studies' below).
**Participant characteristics:** number of participants; gender; parent involved (mother, father, both, unclear); mother's health status; details of inclusion and exclusion criteria, such as previous stillbirths, fetal age and/or reasons for stillbirth; social demographic details including information health literacy; language, ethnicity, age range.
**Intervention:** type of intervention (independent; clinical consultation support; mediated support; unclear); stated aim of intervention; description of informational component including topics and evidence base for information; description of deliberative component; description of other components; other post-mortem investigations offered and their rate of uptake.
**Outcomes:** intervention effect estimate, P value and confidence interval and method of statistical analysis used for all outcomes reported in included studies although our analyses will be confined to those outcomes selected a priori; type of delivery (vaginal delivery, assisted vaginal delivery, elective caesarean, emergency caesarean); decisional conflict; decisional regret; adverse outcomes (distress, conflict); knowledge or understanding of options and possible outcomes; information needs met; satisfaction with decision-making.

We will also record how each outcome was measured and when they were measured. If information about any outcome is unclear, we will contact authors of the original for further details.

### Qualitative data

Due to the lack of a strong theoretical base for the development of decision support interventions, we will examine qualitative studies conducted concurrently with included trials for information related to the acceptability of the intervention and issues related to its feasibility. A narrative synthesis of these issues will be conducted to inform the discussion section of the review. It is anticipated that any qualitative study alongside a trial will include a subset of trial participants and the same inclusion and exclusion criteria will apply.
**Details of study:** study design; description of participants; aim of study; methods of recruitment of participants; differences in inclusion/exclusion criteria for participation in qualitative study and trial; informed consent obtained (yes/no/unclear); ethical approval (yes/no/unclear); funding source and amount (if stated); consumer involvement (in the design of study and interpretation of study findings).
**Participant characteristics:** number of participants; gender; parent involved (mother, father, both, unclear); details of inclusion and exclusion criteria, such as previous stillbirths, fetal age and/or reasons for stillbirth; social demographic details including information health literacy; language, ethnicity, age range.
*Secondary participants: (health professionals, support facilitators):* number of participants; type (doctor, midwife or nurse, pathologist, spouse or partner, other); whether trained in use of intervention (yes/no/unclear); age range; details of inclusion criteria, gender.

## Assessment of risk of bias in included studies

Two review authors will independently assess risk of bias for each study using the criteria outlined in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). We will resolve any disagreement by discussion or by involving a third review author.
(1) **Sequence generation** (checking for possible selection bias)
We will describe for each included study the method used to generate the allocation sequence in sufficient detail to allow an assessment of whether it should produce comparable groups.
We will assess the method as:

- adequate (any truly random process, e.g. random number table; computer random number generator);
- inadequate (any non-random process, e.g. odd or even date of birth; hospital or clinic record number);
- unclear.

(2) **Allocation concealment** (checking for possible selection bias)

We will describe for each included study the method used to conceal the allocation sequence and determine whether intervention allocation could have been foreseen in advance of, or during recruitment, or changed after assignment.
We will assess the methods as:

- adequate (e.g. telephone or central randomisation; consecutively numbered sealed opaque envelopes);
- inadequate (open random allocation; unsealed or non-opaque envelopes, alternation; date of birth);
- unclear.

(3) **Blinding** (checking for possible performance bias)
We will describe for each included study the methods used, if any, to blind study participants and personnel from knowledge of which intervention a participant received. We will consider that studies are at low risk of bias if they were blinded, or if we judge that the lack of blinding could not have affected the results. We will assess blinding separately for different outcomes or classes of outcomes.
We will assess the methods as:

- adequate, inadequate or unclear for participants;
- adequate, inadequate or unclear for personnel;
- adequate, inadequate or unclear for outcome assessors.

(4) **Incomplete outcome data** (checking for possible attrition bias through withdrawals, dropouts, protocol deviations)
We will describe for each included study, and for each outcome or class of outcomes, the completeness of data including attrition and exclusions from the analysis. We will state whether attrition and exclusions were reported, the numbers included in the analysis at each stage (compared with the total randomised participants), reasons for attrition or exclusion where reported, and whether missing data were balanced across groups or were related to outcomes. Where sufficient information is reported, or can be supplied by the trial authors, we will re-include missing data in the analyses which we undertake. We will assess methods as:

- adequate (where 20% or less data for an outcome are missing);
- inadequate (where more than 20% of data for an outcome are missing);
- unclear.

(5) **Selective reporting bias**
We will describe for each included study how we investigated the possibility of selective outcome reporting bias and what we found.
We will assess the methods as:

- adequate (where it is clear that all of the study's pre-specified outcomes and all expected outcomes of interest to the review have been reported);
- inadequate (where not all the study's pre-specified outcomes have been reported; one or more reported primary outcomes were not pre-specified; outcomes of interest are reported incompletely and so cannot be used; study fails to include results of a key outcome that would have been expected to have been reported);
- unclear.

(6) **Other sources of bias**
We will describe for each included study any important concerns we have about other possible sources of bias.
We will assess whether each study was free of other problems that could put it at risk of bias:

- yes;
- no;
- unclear.

The specific sources of bias we will consider for cluster-randomised trials include:
(i) *Recruitment bias*

- adequate (where it is clear that all study participants are recruited to the trial prior to randomisation);
- inadequate (where not all the study participants are recruited to the trial prior to randomisation);
- unclear.

(ii) *Baseline imbalance*

- adequate (where it is clear baseline comparability of clusters, or statistical adjustment for baseline characteristics is reported);
- inadequate (where not all the study participants are recruited to the trial prior to randomisation);
- unclear.

(iii) *Missing cluster data*

- adequate (where it is clear that there are no missing cluster data or loss of individual outcome data);

- inadequate (where clusters or individual outcome data are missing);
- unclear.

(iv) *Statistical analysis*
- adequate (where clustering taken into account);
- inadequate (where clustering is not taken into account);
- unclear.

(7) **Overall risk of bias**
We will make explicit judgements about whether studies are at high risk of bias, according to the criteria given in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). With reference to (1) to (6) above, we will assess the likely magnitude and direction of the bias and whether we consider it is likely to impact on the findings. We will explore the impact of the level of bias through undertaking sensitivity analyses - *see* 'Sensitivity analysis'.

## Measures of treatment effect

### Dichotomous data

For dichotomous data, we will present results as summary risk ratio with 95% confidence intervals.

### Continuous data

For continuous data, we will use the mean difference if outcomes are measured in the same way between trials. We will use the standardised mean difference to combine trials that measure the same outcome, but use different methods.

## Unit of analysis issues

Studies where clusters of individuals are randomised intervention groups (cluster-RCTs, quasi-RCTs), but where inference is intended at the level of the individual, will need to be re-analysed taking account of intra-cluster correlation (ICC) where possible. The design effect will be calculated using the formula 1 + (M -1) ICC , where M is the average cluster size. A common design effect will be assumed across intervention groups. Estimates of ICC will be obtained from contacting authors, or imputed using external estimates from similar studies. If this is not possible, we will report effect estimates and annotate 'unit of analysis error'.
For dichotomous data, both the number of participants and the number experiencing the event will be divided by the design effect and rounded to whole numbers. Small trials will be excluded.
For continuous data, the sample size will be reduced only and means and standard deviations will remain unchanged.

### Cluster-randomised trials

We will include cluster-randomised trials in the analyses along with individually-randomised trials. We will adjust their sample sizes using the methods described in the *Cochrane Handbook for Systematic Reviews of Interventions [Section 16.3.4]* using an estimate of the intra cluster correlation co-efficient (ICC) derived from the trial (if possible), from a similar trial or from a study of a similar population. If we use ICCs from other sources, we will report this and conduct sensitivity analyses to investigate the effect of variation in the ICC. If we identify both cluster-randomised trials and individually-randomised trials, we plan to synthesise the relevant information. We will consider it reasonable to combine the results from both if there is little heterogeneity between the study designs and the interaction between the effect of intervention and the choice of randomisation unit is considered to be unlikely.
We will also acknowledge heterogeneity in the randomisation unit and perform a subgroup analysis to investigate the effects of the randomisation unit.

### Dealing with missing data

For included studies, we will note levels of attrition. We will explore the impact of including studies with high levels of missing data in the overall assessment of treatment effect by using sensitivity analysis.

For all outcomes, we will carry out analyses, as far as possible, on an intention-to-treat basis, i.e. we will attempt to include all participants randomised to each group in the analyses, and all participants will be analysed in the group to which they were allocated, regardless of whether or not they received the allocated intervention. The denominator for each outcome in each trial will be the number randomised minus any participants whose outcomes are known to be missing.

We do not plan to undertake any imputation for missing outcome data other than summary data (ICCs or standard deviations), where possible. We will report all assumptions. We will investigate the affect of our choice of ICCs on the pooled effect estimate in any meta-analysis through sensitivity analyses.

### Assessment of heterogeneity

We will assess statistical heterogeneity in each meta-analysis using the $T^2$, $I^2$ and $Chi^2$ statistics. We will regard heterogeneity as substantial if the $T^2$ is greater than zero and either $I^2$ is greater than 30% or there is a low P value (less than 0.10) in the $Chi^2$ test for heterogeneity.

### Assessment of reporting biases

If there are 10 or more studies in the meta-analysis, we will investigate reporting biases (such as publication bias) using funnel plots. We will assess funnel plot asymmetry visually, and use formal tests for funnel plot asymmetry. For continuous outcomes, we will use the test proposed by Egger 1997, and for dichotomous outcomes, we will use the test proposed by Harbord 2006. If asymmetry is detected in any of these tests or is suggested by a visual assessment, we will perform exploratory analyses to investigate it.

### Data synthesis

We will carry out statistical analysis using the Review Manager software (Revman 2011). We will use fixed-effect meta-analysis for combining data where it is reasonable to assume that studies are estimating the same underlying treatment effect: i.e. where trials are examining the same intervention, and the trials' populations and methods are judged sufficiently similar. If there is clinical heterogeneity sufficient to expect that the underlying treatment effects differ between trials, or if substantial statistical heterogeneity is detected, we will use random-effects meta-analysis to produce an overall summary if an average treatment effect across trials is considered clinically meaningful. The random-effects summary will be treated as the average range of possible treatment effects and we will discuss the clinical implications of treatment effects differing between trials. If the average treatment effect is not clinically meaningful, we will not combine trials.

If we use random-effects analyses, the results will be presented as the average treatment effect with its 95% confidence interval, and the estimates of $T^2$ and $I^2$.

Where multi-armed trials are included, where possible, we will combine groups to create a single pair-wise comparison, i.e. by combining all relevant experimental intervention groups of the study into a single group, and by combining all relevant control intervention groups into a single control group as recommended in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). We will consider selecting one pair of groups for comparison if combining the groups is deemed unacceptable, e.g. if interventions are clinically or statistically heterogeneous. The rationale for this selection will be clearly described in the methods of the review.

### Subgroup analysis and investigation of heterogeneity

If we identify substantial heterogeneity, we will investigate it using subgroup analyses and sensitivity analyses. We will consider whether an overall summary is meaningful, and if it is, use random-effects analysis to produce it.

We plan to carry out the following subgroup analyses.

#### 1. Gestation at birth

- Thirty-three weeks' gestation or more versus up to 24 weeks' gestation and versus 24 to 32 weeks' gestation.

**2. Maternal demographic characteristics**

- Age: 20 to 34 years versus less than 20 years and versus 35 + years.
- Parity: (primiparous versus multiparous).
- Socio-economic status: low versus high.

**3. Country setting**

- Low- and middle-income versus high-income country settings.

**4. Type of interventions**

- Interventions directed at parents versus interventions directed at health professionals.

The following outcomes will be used in subgroup analysis.

- Proportion and type of post-mortem investigations performed.
- Decisional conflict.
- Psychological outcomes (anxiety, depression, etc).

We will assess differences between subgroups by interaction tests available in Revman 2011.

### Sensitivity analysis

Sensitivity analyses will be performed by the quality of included trials excluding studies assessed as having a high risk of bias. We will also undertake sensitivity analysis to explore the effects of fixed- or random-effects analyses for outcomes with statistical heterogeneity and the effects of any assumptions made such as the value of the ICC used for cluster-randomised trials.

## CONTRIBUTIONS OF AUTHORS

Dell Horey wrote the initial version and subsequent drafts of the protocol and review. Dell Horey and Vicki Flenady screened all titles. Dell Horey searched all the websites. All authors contributed to subsequent drafts of the protocol and review. Dell Horey is the guarantor of the review.

## DECLARATIONS OF INTEREST

Dr Alexander Heazell has received a research grant from Sands (UK) to investigate parents' and professionals' views, knowledge and experience of care and counselling for investigations after stillbirth.

## SOURCES OF SUPPORT

### Internal sources

- La Trobe University, Australia.

### External sources

- National Institute for Health Research, UK.

NIHR Programme of centrally-managed pregnancy and childbirth systematic reviews of priority to the NHS and users of the NHS: 10/4001/02

## DIFFERENCES BETWEEN PROTOCOL AND REVIEW

An additional search for information about or reference to autopsy or post-mortem examinations on the member websites of the International Stillbirth Alliance was undertaken.

## INDEX TERMS

### Medical Subject Headings (MeSH)

*Autopsy; *Decision Making; Decision Support Techniques; Parents [*psychology]; Stillbirth [*psychology]

### MeSH check words

Female; Humans; Pregnancy