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Support for mothers, fathers and families after perinatal death (Review)

Koopmans L, Wilson T, Cacciatore J, Flenady V

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[Intervention Review]

Support for mothers, fathers and families after perinatal death

Laura Koopmans[1], Trish Wilson[2], Joanne Cacciatore[3], Vicki Flenady[4]

[1]Mater Medical Research Institute, Mater Health Services, Woolloongabba, Australia. [2]Education and Support Services, Mater Mothers' Hospital, South Brisbane, Australia. [3]School of Social Work, Arizona State University, Phoenix, Arizona, USA. [4]Translating Research Into Practice (TRIP) Centre - Mater Medical Research Institute, Mater Health Services, Woolloongabba, Australia

Contact address: Laura Koopmans, Mater Medical Research Institute, Mater Health Services, Level 2 Quarters Building, Annerley Road, Woolloongabba, Queensland, 4102, Australia. lauskoopmans@hotmail.com, laura.koopmans@mater.org.au.

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ABSTRACT

Background

Provision of an empathetic, sensitive, caring environment and strategies to support mothers, fathers and their families experiencing perinatal death are now an accepted part of maternity services in many countries. Interventions such as psychological support or counselling, or both, have been suggested to improve outcomes for parents and families after perinatal death.

Objectives

To assess the effect of any form of intervention (i.e. medical, nursing, midwifery, social work, psychology, counselling or community-based) on parents and families who experience perinatal death.

Search methods

We searched the Cochrane Pregnancy and Childbirth Group's Trials Register (28 January 2013) and article bibliographies.

Selection criteria

Randomised trials of any form of support aimed at encouraging acceptance of loss, bereavement counselling, or specialised psychotherapy or counselling for mothers, fathers and families experiencing perinatal death.

Data collection and analysis

Two review authors independently assessed eligibility of trials.

Main results

No trials were included.

Authors' conclusions

Primary healthcare interventions and a strong family and social support network are invaluable to parents and families around the time a baby dies. However, due to the lack of high-quality randomised trials conducted in this area, the true benefits of currently existing interventions aimed at providing support for mothers, fathers and families experiencing perinatal death is unclear. Further, the currently available evidence around the potential detrimental effects of some interventions (e.g. seeing and holding a deceased baby) remains inconclusive at this point in time. However, some well-designed descriptive studies have shown that, under the right circumstances and guided by compassionate, sensitive, experienced staff, parents' experiences of seeing and holding their deceased baby is often very positive. The sensitive nature of this topic and small sample sizes, make it difficult to develop rigorous clinical trials. Hence, other research

designs may further inform practice in this area. Where justified, methodologically rigorous trials are needed. However, methodologically rigorous trials should be considered comparing different approaches to support.

PLAIN LANGUAGE SUMMARY

Support for mothers, fathers and families after perinatal death

It is devastating for parents and families when a baby dies. It is estimated that approximately one in five parents will suffer from intense and prolonged grief following the death of a baby around the time of birth. It is essential that parents and families are offered, and have access to, appropriate support from caregivers and their direct social network. Yet, little is known about the role and the true effectiveness of different types of bereavement support for parents and their families. This review aimed to identify clinical trials to assess the effect of different types of bereavement support interventions and/or counselling for parents experiencing perinatal death. There are no included studies on this topic. For the update of this review we identified one new trial, which is currently awaiting classification. More research in this area is needed.

BACKGROUND

The death of a child around the time of birth is one of the most profound, stressful events an adult may experience (Bonanno 2001; Fish 1986; Wing 2001). For decades, mothers (and fathers) were separated from their stillborn or dying babies in the belief that grief could be prevented if no attachments were formed. After it was established that attachment relationships between mother and child are already formed during pregnancy, (Giles 1970; Kennell 1970), research has focused on exploring the substantial impact of perinatal death on parents and families.

Grief reactions

Normal parental grief reactions immediately following perinatal death have been well documented and resemble those in other bereavement situations (e.g. after the death of a spouse). Profound sadness, depressed mood, irritability, preoccupation, anxiety and changes in eating and in sleeping patterns are all considered to be part of a normal grief response (Burnett 1997; Parkes 1972; Raphael 1984). Symptoms of acute grief typically subside with time and for most people the intensity has significantly reduced by six to 12 months post-loss (Bonanno 2001; Shear 2011). Yet, grief recovery or rather 'the normalisation of the psychosocial effects of perinatal death' has been reported to take as long as five to 18 years (Gravensteen 2012).

Pathological responses to bereavement include bereavement-related major depression, post-traumatic stress disorder (PTSD) and complicated grief (Lichtenthal 2004; Stroebe 2008a) and it is not uncommon that these conditions co-occur in bereaved individuals (Shear 2005). Pathalogical grief responses are more likely to occur in patients with a pre-existing mental health diagnosis. Although the majority of bereaved parents will experience normal grief, bereaved parents have been repeatedly identified to be at increased risk of complicated grief (Badenhorst 2007; Hughes 2003; Korenromp 2007; Radestad 2009). Persisting and significant grief-related problems may be more prevalent in a subset of parents. Kersting *et al* (2007) found that recently bereaved mothers who had a termination of pregnancy (TOP) for fetal abnormalities, were significantly more likely than controls to suffer from a range of psychiatric disorders for up to 14 months after their loss (Kersting 2007). In this cohort, acute stress disorders such as PTSD, were mostly resolved at 14 months post-loss, while anxiety and affective disorders were the most common diagnoses at the 14-month point.

Post-traumatic stress has also been reported in the subsequent pregnancy following perinatal death. Although a clinical diagnosis of PTSD was not made, one study (Turton 2001) reported that 20% of women fulfilled the criteria of PTSD during a pregnancy following stillbirth, compared with the general PTSD population incidence of 5% to 10% (Keane 2009). One year postpartum, however (i.e. following the birth of a healthy baby), both current and lifetime PTSD rates in these women had decreased to around 5%, similar to population levels. At seven-year follow-up, there were no longer significant differences in PTSD and major depression between bereaved mothers and controls.

Relationships

Perinatal death has been identified as a risk factor for relationship break-down (Najman 1993; Vance 2002). Gold and colleagues found that stillbirth increased the risk of parental separation by 40% (adjusted Hazard Ratio (aHR) 1.40; 95% confidence interval (CI) 1.10 to 1.79) (Gold 2010). Similarly, Shreffler *et al* found that women who had experienced stillbirth had a significantly increased risk of divorce post-loss (Odds ratio (OR) 1.70; $P < 0.05$) (Shreffler 2012). Another study found that for couples with a previous stillbirth, the risk of relationship breakdown was fourfold compared with couples with no history of stillbirth (OR 4.3; 95% CI 1.6 to 12.0) (Turton 2009). However, this study did not control for important relationship factors, which may partly explain the larger effect of stillbirth on the risk of relationship breakdown.

Perceived partner support after the death of a loved one is well known to be a significant protective factor against lasting grief and distress (Buchi 2009). Couples who share and communicate their grief report less severe grief reactions and greater partner satisfaction (Buchi 2009; Kamm 2001). This suggests that congruent grieving within couples leads to better relationship outcomes and, conversely, that incongruent grief could result in relationship problems. A small, unique study investigated this concept and found that emotional exchange between parents was reflected by concordant grieving in which levels of suffering, depression and anxiety as well as processes of post-traumatic growth were shared by parents. In contrast, parents with disconcordant grief were also disconcordant in suffering, depression and anxiety and did not share post-traumatic growth. Separate, independent experiences of grief, suffering and post-traumatic growth are likely to negatively impact relationship dynamics and satisfaction and may even result in separation (Buchi 2009). Based on the concept of grief concordance, it is not surprising that despite intensified relationship stress, some couples indicate that their loss has “brought them closer together” increasing the relationship cohesion (Cacciatore 2008a; DeFrain 1990).

Fathers

When a baby dies, mothers generally report more severe and enduring grief than fathers (Murray 2000). However, the more active parenting role of today's fathers is likely to impact on grief intensity. Increased prenatal attachment associated with modern obstetric practices, such as prenatal diagnostic procedures, assisted reproduction and graphic ultrasound imaging, has been reported to increase the intensity of mothers' grief (Robinson 1999); it is therefore reasonable to expect that fathers too may experience more intense grief with increasing attachment.

Research indicates both similar and distinctly different grief responses in mothers and fathers after perinatal death. A review of the effects of perinatal death on fathers (Badenhorst 2006) identified common themes in paternal and maternal grief such as shock, anger, emptiness, helplessness and loneliness. Feelings of guilt were frequently reported by mothers but were rarely reported by men. Although findings on maternal and paternal responses to perinatal death are relatively consistent across studies and provide useful information, studies tend to lack statistical power and design quality. Hence, well-designed studies which take a more systematic approach to identifying affective and behavioural responses that are specific to mothers and fathers are needed.

Interventions

The narrative review by Forrest 20 years ago (Forrest 1989) on support after a perinatal death highlighted the need for further

high-quality research in this area. Over time, an abundance of studies have been conducted in the area of perinatal loss, leading to the development of clinical practice guidelines and the widespread development and implementation of a range of support interventions. Common interventions described in the literature include a wide range of medical and psychosocial interventions, provided in both the antenatal and postnatal period.

Prenatal genetic testing

The death of a baby can occur at any time during the perinatal period, the first of which is during pregnancy. With over 500 prenatal genetic tests currently available and increasingly sophisticated ultrasonography, parents can be informed of the potential risk or diagnosis of a fetal abnormality as early as the first or second trimester of pregnancy. In order to make informed decisions about whether or not to continue the pregnancy, parents need to be provided with clear, unbiased information and receive continuous, compassionate guidance and support throughout the various stages of testing and the decision-making process (Scully 2007).

Termination of pregnancy (TOP)

Most studies of women who undergo TOP for fetal abnormalities report significant grief within the first four to six months. Although the majority of these women will adapt and recover well, this group has been identified to be at increased risk of complicated grief (Kersting 2004; Korenromp 2005; Zeanah 1993). Recommendations for care include better information and preparation of those women and the development of specific, relevant grief therapy interventions. Physicians and other healthcare providers should inform and prepare women that their loss may have a significant, long-lasting impact (Kersting 2006).

Palliative care

Perinatal palliative care (PNPC) is an emerging field within the area of perinatal loss which aims to provide care for dying babies and their parents. For some couples it can provide an alternate option to TOP for fetal abnormalities (Breeze 2007). To date, no empirical studies have determined the best model for perinatal palliative care, however, a small number of clinical studies have identified key components to be: early engagement, continuity of antenatal care, a family-centred approach to care, and multi-disciplinary team involvement, including a bereavement specialist. Centres that provide a perinatal hospice or palliative care service report up to 87% uptake (Balaguer 2012).

Supporting parents in end-of-life decision-making when a baby is dying or when continuation of care is futile, includes clear, compassionate communication, physical and emotional care, collaborative decision making and follow-up care (Williams 2008). Active parental involvement in discussions and decisions about withholding or withdrawing care has not been found to aggravate or prolong parental grief or increase the incidence of grief pathology (Schulze 2007). A model of shared decision-making based on the discussion of mutual 'goals of care' gives equal weight to the family and the medical team and often reduces the potential for conflict (Schulze 2007).

Effective neonatal pain and symptom management, discussing options for parental involvement in the baby's dying, preparation for the death and guided decision-making are important aspects of palliative care (Armentrout 2009; Kaempf 2009; Munson 2007). Common challenges to providing comprehensive end-of-life care include care-giver comfort, consistency of care, cultural and legal barriers, and lack of adequate staff training (Kain 2006).

Birthing options after diagnosis of fetal death

Following the diagnosis of an intrauterine fetal death (IUFD), most women opt for delivery of the baby within 48 hours (Silver 2010). The timing of delivery depends on a variety of factors and management should be individualised, however, postponing the birth too long may increase maternal psychological distress and anxiety. One study found that women who postponed delivery for more than 24 hours, had a fivefold increased risk of long-term anxiety related symptoms (Radestad 1996).

Mode of birth is largely dependent on fetal gestational age and maternal clinical history, with consideration of the couple's personal preference. Induction or augmentation of labour and natural vaginal birth has the lowest medical risk for women (Villar 2007). Caesarean section should be reserved for women when clinically indicated. Although couples with a diagnosis of IUFD commonly consider a caesarean section as their initial preferred mode of birth (Samuelsson 2001), women with no clinical indication for this procedure should be encouraged to consider a normal labour due to the known increased risks associated with previous caesarean section in a subsequent pregnancy (Flenady 2011b; Lydon-Rochelle 2001; O'Neil 2013). Reassurance should be given to the parents that pain relief and physical and emotional support during labour and birth will be provided.

Pain relief and sedation

A systematic review on hospital care after perinatal death found that pain relief is often inadequate (Gold 2007a). Yet, sedation is often over-prescribed after perinatal loss (Harper 1994). A large anonymous national survey of obstetricians in the United States revealed that 48.5% supported the prescription of sedatives for a grieving mother in acute bereavement care (Gold 2008), despite the lack of evidence for its benefit in the improvement of sleep or grief (Warner 2001). Furthermore, a significant body of evidence exists about the potential addictive nature of this group of medications. It is strongly recommended that pharmacological management of grief should only be considered in the presence of an established psychological disorder for which medication is indicated (Raphael, Minkov et al. 2001).

Seeing and holding

Currently, most best practice guidelines recommend that all parents should be offered a choice about whether or not they want to see and hold their stillborn baby, and that parents should be supported throughout this process (Flenady 2009; NICE 2010; SANDS 2010). However, the evidence around the benefit of holding and seeing remains somewhat controversial, providing no simple directions to guide staff and parents.

A controversial study in the UK of mothers with a subsequent pregnancy following a stillbirth found that seeing and holding a stillborn baby was associated with worse maternal psychological outcomes (Hughes 2002). Mothers who saw their stillborn baby were more likely to experience anxiety in the third trimester of a subsequent pregnancy. By one year postpartum, anxiety had resolved but PTSD symptoms were higher compared with women

who had not seen their baby. Women who held their stillborn baby were more likely to experience symptoms of PTSD in the third trimester of a subsequent pregnancy and one year after delivery, but this was not the case for depression or anxiety. Follow-up of the mothers at seven years indicated that higher rates of PTSD symptomatology persisted over time (Turton 2009). The study by Hughes and colleagues has been heavily criticised by both bereaved parents and researchers who feel that the translation of the study results to all women should be made with caution due to issues with cohort representation, small sample size and the lack of detail provided around how women were presented with (the option of seeing or holding) their stillborn baby (Ambuehl 2002; Brooks 2002; Kersting 2002; Matthews 2002; McCabe 2002). Methodological shortcomings of the study, including sample size and inclusion of women in a subsequent pregnancy only who had no other living children, limits the generalisability of the study findings.

In a study on late stillbirths (greater than 28 weeks gestation), nearly all mothers who held their stillbirth baby found the experience valuable (Radestad 2009). Mothers who felt they had not received enough support from hospital staff to hold their baby were four times more likely to have not held their stillborn baby when compared with mothers who felt supported. The importance of perceived staff support and attitudes in influencing parental decision-making about seeing and holding a deceased baby is well documented (Radestad 2009; Ransohoff-Adler 1989; Trulsson 2004).

Anecdotal evidence suggests that bereaved parents have much appreciated the experience of seeing and holding their stillborn baby. Women who experienced a stillbirth in the past have repeatedly come forward in more recent years, expressing their distress about not being allowed to see or hold their stillborn baby at the time and not knowing what happened to their babies' bodies. Despite a lack of empirical evidence, research and opinion papers published on this topic generally agree that holding and seeing a stillborn baby is valuable for most, but not all women, and that staff should hence be mindful and sensitive to the individual needs and wishes of each family (Baker 2009; Radestad 2009; Sloan 2008).

Memory creation

Activities that support parents in developing a bond with their baby help create a sense of identity of the child (Klass 1996; Klass 1999). Clinical guidelines support activities such as bathing and dressing the baby, talking to the baby and using the baby's name, engaging in religious or naming ceremonies, introducing the baby to extended family, and capturing interactions in photographs and movies. For many parents, it is the experience of parenting, not mementos, which is the most valuable in the creation of a bond.

There is general consensus that bereaved parents should be offered items of memorabilia such as photos, hand/footprints and special clothing or blankets when a baby dies (Gold 2007a; Henley 2008; Radestad 1996; Silver 2010). The collecting of such items does not appear to lead to adverse grief outcomes (Hughes 2002) and not having such items has been linked to increased anxiety in mothers of stillborn babies (Radestad 1996). A meta-analysis of hospital care for parents after a perinatal loss found that parents overwhelmingly appreciated having photos and memorabilia of their deceased baby, and frequently expressed regret if these were not provided by the hospital (Gold 2007a). Fathers reported that tokens of remembrance were invaluable, and were appreciative of staff collecting them, even if they were declined (Radestad 1996; Samuelsson 2001).

Parent information resources and web-based support

One case-controlled study has demonstrated the benefit of specially designed perinatal grief resources for bereaved families (Murray 1999). Resources included children's story books, and individualised parent information and staff brochures. Web-based mental health services, including informative websites, online self-help groups, virtual counselling services and automated therapy programs for specific mental health problems such as post-traumatic stress (Knaevelsrud 2007; Lange 2003) and complicated grief (Wagner 2006), have emerged more recently and may be able to offer useful support options for some parents experiencing perinatal death (Kersting 2009; Kersting 2011a).

Kersting *et al (* Kersting 2011a) designed, trialed and assessed an online cognitive behavioural therapy protocol adapted from Wagners Internet-based program for clients suffering from complicated grief, and adjusted it to meet the needs of adult mothers who had recently experienced pregnancy loss (i.e. miscarriage, TOP for fetal anomalies or stillbirth). Compared to controls, the cognitive behavioural therapy group, consisting of mothers who did not have serious mental health problems prior to commencing online cognitive behavioural therapy, showed significant improvements in post-traumatic stress (P = 0.012), grief intensity (P = 0.001) and overall mental health (P = 0.004).

Online support groups and memorial websites have become popular in recent times. It is frequently assumed that such resources can be a useful source of psycho-education and provide a sense of emotional and appraisal support (Glanz 2008) for bereaved individuals, however scientific investigation to back up such claims falls far short of acceptable standards. The potential for damage is frequently neglected, with concerns for exploitation or abuse of the lonely and vulnerable, and the possibility that Internet activity reduces socially interaction, potentially reducing much needed social support for the bereaved. With no randomised controlled trials or criteria or procedures to differentiate high-quality from low-quality Internet resources, the value of such resources cannot be determined, and caution should be taken with recommendation (Stroebe 2008b).

Social support

The social environment of the griever has been identified as a significant factor in grief outcomes (Doka 1999), and the role of social support in parental grief has been well documented (Hutti 2005; Umphrey 2011; Zeanah 2006). Qualitative studies demonstrate a correlation between support (from doctors, nurses and families) and lower levels of anxiety and depression in mothers following a stillbirth, with family support reported as most significant (Cacciatore 2008b). Support from partners, family and those outside the family has been shown to reduce maternal distress in the long-term (15 months), though not in the short-term (Murray 1999). The role of support groups in perinatal loss is less clear. A few qualitative studies report a range of important benefits, particularly for women (Cacciatore 2007). Despite the potential value for some mothers however, without well-designed studies to measure both qualitative and quantitative outcomes, support groups may not be recommended for all grieving mothers.

Culturally sensitive care

The importance of recognising the cultural perspective of loss and grief is well supported in the adult loss and grief literature (Butler 2012; Stroebe 1998; Walter 2010), however only limited attempts have been made to explore the cultural context of perinatal loss. Seminars in Fetal and Neonatal Medicine (October 2008) published a series of discussion papers on cultural perspectives of care in foetal and neonatal medicine, (Evans 2008; Gatrad 2008; Husain 2008; Laing 2008; McGraw 2008; Nelson 2008; Rennie 2008; Shinwell 2008; Steer 2008; Vaughan 2008; Williams 2008). They conclude the importance of sensitive cultural approaches and encourage further research in this area of perinatal care. Others (Chichester 2005; Laing 2008) caution imposing a 'Western grief culture' which values engagement with death and grief onto other cultures. Staff's knowledge and understanding of key religious and cultural rituals can greatly facilitate difficult discussions and decision-making around the time of death of a baby (Gatrad 2008).

Counselling and psychotherapy

No robust studies have been undertaken in the area of perinatal loss to determine the effect of grief counselling or psychotherapy on parental grief. A recent systematic review of 61 controlled outcome studies of grief counselling in the general bereaved population over the last three decades found only small observed advantages in treated clients compared to untreated controls, effects which are lost over time (Neimeyer 2010). Authors conclude that universally applied bereavement interventions do not achieve measurable benefit when compared with 'no treatment' groups, and the majority of grievers experiencing 'normal' grief will adapt to their loss or respond resiliently (Bonanno 2004; Neimeyer 2010). This is in contrast to high-risk groups such as parents who have lost children, (Neimeyer 2010, p6) or grievers with significant symptomatolgy, such as those with complicated grief or clinical depression, who do receive benefit. The task for primary clinicians in the area of parental bereavement will be to identify parents who are at increased risk of pathological grief, and who would benefit from referral to mental health services.

There is no doubt that compassionate, sensitive care is invaluable for bereaved parents and families (Janzen 2003-2004; Kirkley-Best 1982; Mashegoane 1999; Murray 2000; Wing 2001). The importance of appropriate psychosocial support for all women and families globally was recently highlighted in a comprehensive international stillbirth series published in the Lancet (Froen 2011; Flenady 2011a, as part of www.thelancet.com/series/stillbirth [Lancet Stillbirth series 2011]). Yet, what continues to pose difficulties for those attempting to provide perinatal bereavement support is exactly what it is that comprises 'best practice', particularly in relation to psychosocial care. We undertook this review to identify evidence from high quality randomised controlled trials, looking at optimal approaches for supporting parents and families experiencing perinatal death.

OBJECTIVES

The specific objectives of this review are to determine the effectiveness of any form of medical, midwifery, nursing, psychological or social support in preventing or reducing the incidence or severity, or both, of (protracted) grief reactions or long-term psychopathological sequelae, or both, in mothers, fathers and families experiencing perinatal death.

METHODS

Criteria for considering studies for this review

Types of studies

Randomised controlled trials were considered for inclusion in this review if:

- perinatal death was defined as stillbirth or neonatal death according to the definitions used in each trial;
- the study compared any social or professional support, or both, after perinatal death, with standard care as practiced at the time of the study;
- the proportion of loss to follow-up was no more than 30%.

Types of participants

Mothers and/or fathers and/or their immediate families, experiencing the death of a baby in the perinatal period. Trials involving early spontaneous pregnancy losses (i.e. spontaneous miscarriages before 20 weeks' gestation or as according to the definition of miscarriage used in each trial) or termination of pregnancy (TOP) for non-medical reasons were excluded.

Types of interventions

Any type of intervention provided by professional or non-professional individuals or groups which are aimed at improving the psychological well-being of parents and families after perinatal death. These may include any form of:

- general supportive hospital interventions aimed at supporting parents around the time of their baby's death. This may include parent information provision after a stillbirth diagnosis or diagnosis of a fetal abnormality, photographs and other memorabilia, holding and naming the baby, offering dignified funeral rites or disposal arrangements for stillbirths, and hospital follow-up visits;
- specific religious, spiritual and/or cultural supports;
- interventions labelled as bereavement counselling;
- specialised psychotherapy, counselling, or assessment, either single or multiple sessions or therapeutic episodes;
- interventions for women with a previous perinatal death in the subsequent pregnancy;
- community and online support groups.

Types of outcome measures

Primary outcomes

The primary outcome measures include:

- normal grief reactions including depressed mood and anxiety;
- pathological grief reactions, including post-traumatic stress and complicated grief;
- satisfaction with care.

Secondary outcomes

Secondary outcome measures include:

- physical symptoms of grief;
- signs of social maladjustment;
- family disruption; relationship disharmony or breakdown;

- cost of interventions.

Where appropriate, these outcomes are definable by standard clinical criteria and measurable by standard psychometric methods such as questionnaires or interviews, or both. Where possible, we planned that subgroup analyses of outcomes for high-risk groups would be conducted (i.e. women with TOP for fetal abnormality, poor support and subsequent pregnancy).

Search methods for identification of studies

Electronic searches

We searched the Cochrane Pregnancy and Childbirth Group's Trials Register by contacting the Trials Search Co-ordinator (28 January 2013).

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; and
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.

Searching other resources

We searched reference lists of retrieved articles.

We did not apply any language restrictions.

Data collection and analysis

For the methods used when assessing trials identified in the previous version of this review, see Appendix 1.

Selection of studies

Two review authors independently assessed for inclusion all the potential studies we identified as a result of the search strategy. We planned to resolve any disagreement through discussion or, if required, we planned to consult a third person.

When information regarding any of the above was unclear, we contacted authors of the original reports to provide further details.

In future updates, we will use the methods for assessing eligibility, data extraction and management, risk of bias, and data synthesis described in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011) and set out in detail in Appendix 2.

RESULTS

Description of studies

One new trial was identified (Kersting 2011b). However, the data format reported in this trial did not allow us to separate out treatment outcomes for women who had a stillbirths or late TOP for fetal abnormalities from women who had miscarriages or early TOP. The author of the trial was contacted with the request to kindly provide, if possible, study findings specific to women who had a stillbirth or late TOP for fetal abnormalities. Pending the response, this trial is currently awaiting classification for the purposes of this review (see table Characteristics of studies awaiting classification).

In the previous version the authors identified three potentially eligible trials (Forrest 1982; Lake 1987; Lilford 1994) but these were all excluded (*see* table Characteristics of excluded studies). The large loss to follow-up rate was the major reason for exclusion.

Risk of bias in included studies

Not applicable.

Effects of interventions

Not applicable.

DISCUSSION

This review has highlighted the difficulty of research in the area of grief support around the time of perinatal death, and the ongoing lack of empirical evidence that has arisen from existing studies. From being largely neglected in the past, it is encouraging to see some randomised controlled trials being undertaken to address the lack of high level evidence. However, the available empirical data remains sparse and variable, and trials are of insufficient quality, size and comparability to enable any truly valid conclusions.

One of the excluded trials (Lake 1987) was set in a population of predominantly indigent single or poorly socially-supported mothers, or both, in west central Florida, while the other two (Forrest 1982; Lilford 1994) were set in large British teaching hospitals and included partners in the intervention. The most recent trial (Lilford 1994) was the only one that included couples who had experienced termination of pregnancy (TOP) for fetal anomaly as well as stillbirths and neonatal deaths.

The three excluded trials (*see* table Characteristics of excluded studies) do however provide some insight into the areas of difficulties in these studies, and may guide the design of future trials. Only one trial (Lilford 1994) provided power calculations of the numbers needed to be randomised, and the results of this trial could be utilised towards better estimation of the numbers required in future studies to retest the hypothesis. The large loss to follow-up rate was the major reason for exclusion of all three trials, and should alert future researchers to specifically target this problem and to seek sufficient resources to enable better follow-up.

All three trials identified certain high-risk groups that may warrant further study. Two (Forrest 1982; Lake 1987) noted that socially-isolated women or women with low levels of social support tended to have a higher incidence of psychiatric symptoms. One trial (Lilford 1994), suggested that women who underwent TOP for fetal anomalies had slightly worse outcomes than those who had

experienced stillbirth or neonatal death. This is likely to be related to the specific grief issues related to TOP, including active decision making, guilt and shame. Given the difficulty of research in this area, it may be that specific emphasis and attention to these high-risk groups, with adequate levels of follow-up, may be warranted.

Although two trials (Forrest 1982; Lilford 1994) included partners in the interventions, they were not able to draw any specific conclusions and further attention to the effects of such interventions for fathers is needed.

The influence of cultural and racial differences on the incidence of psychiatric symptoms remains a potentially interesting but unexplored aspect of adjustment to perinatal death and as yet no randomised controlled trials have specifically addressed this issue.

The current discussion and planning towards the inclusion of complicated or pathological grief as a distinct category of mental disorder in the upcoming Diagnostic and Statistical Manual of Mental Disorders (DSM-V) will have implications on both the identification and potential interventions for bereaved parents (Zhang 2006). Clarity of the definition and classification of pathological grief is likely to increase the identification of a population for whom intervention is likely to be helpful, and this in turn is likely to lead to the development of interventions that can be empirically tested.

Another emerging area of research that will contribute to our scientific knowledge of grief interventions is that of the neurobiology of grief and trauma indicating that grief is mediated through a neural network across regions of the brain (Gündel 2003).

Also, growing research into resilience (Zhang 2006) and post-traumatic growth (Buchi 2007) will add further to the development of effective post-loss interventions.

AUTHORS' CONCLUSIONS

Implications for practice

Due to the lack of randomised trials in this area, this review cannot provide clear guidance for best practice in the area of support for parents and families following a stillbirth or neonatal death. Nontheless, providing support for parents and families after perinatal death is justified based on the study findings of non-randomised studies discussed in this review, and the authors have highlighted a range of interventions that may be useful to parents and families. It is evident from this review that three themes are consistent in providing care for bereaved parents: firstly a deep respect for the individuality and diversity of grief, respect for the deceased child, and recognition of the healing power and resilience of the human spirit.

Implications for research

Methodologically rigorous trials are needed in order to assess the true effects of interventions aimed at providing appropriate and sensitive support for parents and families after a perinatal death. Certain high-risk groups (women with pre-existing mental health issues, women with termination of pregnancy) may need to be specifically targeted, as will the effect of interventions for fathers. It is likely that multi-centre studies will be necessary, with adequate funding to ensure proper follow-up in order to definitively address these questions. Further, trials should ensure that the range of outcome measures is clearly defined and is assessed by standard psychometric tools, as far as possible validated for the purpose, that data are numerically complete and appropriately presented, and that adequate follow-up is possible.

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CHARACTERISTICS OF STUDIES

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

Study	Reason for exclusion
Forrest 1982	High loss to follow-up, particularly in treatment group. At 6 months, the loss to follow-up in the treatment group was 36% and in the control group, 24%. 25 women were randomised to the treatment group and 25 to the control group.
Lake 1987	Overall high loss to follow-up. At 6 months, 44 women (56.4%) of the 78 women recruited were lost to follow-up. Randomisation method not stated. Data available are in an unsuitable form for analysis.
Lilford 1994	High loss to follow-up. At 16-20 months, the loss to follow-up for women enrolled in the study was 51.4% for the treatment group (N = 18/57) and 6.3% for the control group (N = 8/57). Randomisation method was stated. Strong possibility of selection bias (22 randomised to control group, 35 to treatment group). Data available are in an unsuitable form for analysis.

Characteristics of studies awaiting assessment *[ordered by study ID]*

Kersting 2011b

Methods	Randomised controlled trial
Participants	83 German speaking mothers who had lost a child during pregnancy through miscarriage, termination of pregnancy due to fetal anomaly, or stillbirth
Interventions	5-week internet-based cognitive behavioural therapy versus 5-week waiting condition
Outcomes	Relative to controls, participants in the treatment group showed significant improvements in post-traumatic stress, grief, depression, and overall mental health, but not in anxiety or somatization. Medium-to-large effect sizes were observed, and the improvement was maintained at 3-month follow-up. Stillbirth data were not separately reported.

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Kersting 2011b *(Continued)*

Notes	Annette Kersting, the lead investigator of this trial, was contacted by the authors of this review on 11 April 2013, with the request to kindly provide data specific to women who had a stillbirth.

APPENDICES

Appendix 1. Methods used to assess trials included in previous versions of this review

Two review authors (Helen Chambers and Fung Yee Chan) independently selected the trials to be included in the orginal version of this review (Chambers 1998) with the reasons for exclusion of any apparently eligible trial clearly stated. Any disagreement was resolved by discussion. The same review authors assessed the methodological quality of the trials with details of randomisation, blinding and exclusions from the analyses recorded.

Review author (Vicki Flenady) subsequently assessed the quality of the identified trials as a part of the 2008 update of the review (Flenady 2008).

We attempted to contact trial authors for additional information to allow both assessment of methodological quality and to permit 'intention-to-treat' analysis of data. Dr Gillian Forrest and Professor Richard Lilford provided additional information about their published trials.

Appendix 2. Methods for use in future updates

Selection of studies

In future updates of this review, two review authors will independently assess for inclusion all the potential studies we identify as a result of the search strategy. We will resolve any disagreement through discussion or, if required, we will consult a third person.

Data extraction and management

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 review author. We will enter data into Review Manager software (RevMan 2012) and check for accuracy.

When information regarding any of the above is unclear, we will attempt to contact authors of the original reports to provide further details.

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 assessor.

(1) Random 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:

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

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

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

We will assess the methods as:

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

(3.1) Blinding of participants and personnel (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 would be unlikely to affect results. We will assess blinding separately for different outcomes or classes of outcomes.

We will assess the methods as:

- low, high or unclear risk of bias for participants;
- low, high or unclear risk of bias for personnel.

(3.2) Blinding of outcome assessment (checking for possible detection bias)

We will describe for each included study the methods used, if any, to blind outcome assessors from knowledge of which intervention a participant received. We will assess blinding separately for different outcomes or classes of outcomes.

We will assess methods used to blind outcome assessment as:

- low, high or unclear risk of bias.

(4) Incomplete outcome data (checking for possible attrition bias due to the amount, nature and handling of incomplete outcome data)

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 and 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:

- low risk of bias (e.g. no missing outcome data; missing outcome data balanced across groups);
- high risk of bias (e.g. high attrition (greater than 20%) or reasons for missing data imbalanced across groups; 'as treated' analysis done with substantial departure of intervention received from that assigned at randomisation);
- unclear risk of bias.

(5) Selective reporting (checking for 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:

- low risk of bias (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);
- high risk of bias (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 risk of bias.

(6) Other bias (checking for bias due to problems not covered by (1) to (5) above)

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:

- low risk of other bias;
- high risk of other bias;
- unclear whether there is risk of other bias.

(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* (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

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* using an estimate of the intracluster 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 [*sensitivity OR 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.

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 an I^2 is greater than 30% and either a T^2 is greater than zero, 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. If asymmetry 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 2012). 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 of the 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 95% confidence intervals, and the estimates of T^2 and I^2.

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. Type of intervention: person delivering the intervention: clinician (midwife, obstetrician); professionals specifically trained in bereavement counselling; qualified psychologist/psychotherapist, other professionals (e.g. social workers, pastoral care workers), or other (e.g. bereaved parents).
2. Mode of delivery and intensity of the intervention: e.g. face-to-face, telephone or internet-based interventions, and the duration of time that specific support was provided (i.e. number of consultations).
3. High-risk population: interventions provided for parents or families, or both who are considered to be at increased risk, namely: 1) termination of pregnancy for fetal anomalies (TOP), 2) parents with previous mental illness, 3) complicated grief.

We will carry out subgroup analysis for primary outcomes only.

We will assess subgroup differences by interaction tests available within RevMan (RevMan 2012). We will report the results of subgroup analyses quoting the $\chi 2$ statistic and P value, and the interaction test I^2 value.

Sensitivity analysis

We will carry out sensitivity analysis: if we include trials at high risk of bias, we will temporarily remove them from the analysis, to examine the impact of excluding these trials; if we include cluster-randomised trials, we will examine the effect of varying the ICC.

F E E D B A C K

Lang, September 2005

Summary

The authors are to be commended for taking on the important, and often neglected, issue of providing support to bereaved families following perinatal loss.

None of the published studies met the quality criteria for inclusion in the review, and data on this topic are described as 'sparse' and 'variable'. In their discussion, the authors appropriately identify the limitations of their study, but then go on to state that the lack of trials was further complicated by "the evolution over 15 years of so called 'standard care' after perinatal death: the provision of an empathic caring environment, which was regarded in the earlier trials as part of the intervention, is now standard care in most centres". The basis for this conclusion is, however, questionable, and there is evidence to the contrary. Indeed, among health professionals there continues to be a sense of discomfort with the subject matter that frequently spills over into the care provided, which is often inadequate and can actually be detrimental.

Conclusions emanating from reviews where no quality studies are included must be carefully considered, and should be well-substantiated by other evidence. Ill-informed conclusions cited in *The Cochrane Library* can have an important impact on practitioners, researchers and funders.

(Summary of comments from Ariella Lang, September 2005)

Reply

We thank Ariella Lang for her comments and hope that our reply adequately addresses the concerns raised regarding our comments in the discussion of the review on the quality of current practice for parents after a perinatal death.

We agree that care for parents around the time of a perinatal death often falls short. We also agree that a sense of discomfort by healthcare professionals when dealing with a perinatal death may have negative effects on the quality of care and outcomes for parents. To better reflect this, the issue of care around the time of death is now discussed with appropriate references in the background, and the sentences about evolution of care have been removed from the background and discussion. Also, the list of interventions included in the review has been expanded to include support and education for professionals on perinatal bereavement. However, for this update we were not able to identify any randomised trials addressing this intervention.

The conclusions of the review clearly highlight the current lack of evidence to guide care and the need for well-designed trials to determine the appropriate support interventions for parents following a perinatal death. As is discussed in the conclusion, this lack of clearly defined and tested interventions may affect the confidence of practitioners, as well as funding opportunities, which may further contribute to the inadequate care currently provided to families who experience perinatal loss.

(Summary of reply by Vicki Flenady and Trish Wilson, May 2007)

Contributors

Feedback: Ariella Lang
Response: Vicki Flenady, Trish Wilson

WHAT'S NEW

Date	Event	Description
15 March 2013	New citation required but conclusions have not changed	No new trials included. One new trial currently awaiting classification (Kersting 2011b).
28 January 2013	New search has been performed	Search updated. Methods updated.

HISTORY

Protocol first published: Issue 3, 1997
Review first published: Issue 2, 1998

Date	Event	Description
23 November 2011	New search has been performed	Search updated and no new trials identified. Plain language summary added. Background modified.
11 November 2010	New search has been performed	List of contributing authors updated
29 January 2009	Amended	Author's contact details edited.
11 February 2008	Amended	Converted to new review format.
6 November 2007	New citation required and conclusions have changed	Substantive amendment

CONTRIBUTIONS OF AUTHORS

For this update, Laura Koopmans and Trish Wilson revised the background and discussion of the previous version of this review, in collaboration with Vicki Flenady and Joanne Cacciatore.

DECLARATIONS OF INTEREST

None known.

SOURCES OF SUPPORT

Internal sources

- Mater Medical Research Institute and the Mater Mothers' Hospitals, Mater Health Services, Brisbane, Australia.

External sources

- NIHR NHS Cochrane Collaboration Programme grant scheme award for NHS-prioritised centrally-managed, pregnancy and childbirth systematic reviews: CPGS 10/4001/02, UK.

INDEX TERMS

Medical Subject Headings (MeSH)

*Bereavement; *Counseling; *Death; *Nuclear Family; *Social Support; Life Change Events

MeSH check words

Humans; Infant, Newborn