Contact with the stillborn baby and parental mental health and wellbeing: a systematic review

Protocol

Julie M Hennegan (JMH)
Jane Henderson (JH)
Maggie Redshaw (MR)
Background

Stillbirth occurs in around 1 in 200 pregnancies in high-income countries (Flenady et al., 2011). Perinatal loss is a devastating and traumatic event for women and their partners. The grief and distress experienced by parents has been documented across qualitative and quantitative studies (Turton, Hughes, Evans & Fainman, 2001; Turton, Evans & Hughes, 2009; Harper, O’Connor & Carroll, 2011; Keeley & Trinidad, 2012; Mills et al., 2014). This bereavement, however, has often been misunderstood, stigmatised and failed to receive adequate recognition (Froen et al., 2011; Cacciatore, 2013). Recent investigation of parents’ experiences in the UK has emphasised the importance of events and care provided at this sensitive time, highlighting that everyone involved had ‘only once chance to get it right’ (Downe, Schmidt, Kingdon & Heazell, 2013).

Standard practices in caring for parents during and after stillbirth have varied over time. Traditionally parents did not see or hold their baby and the stillborn was taken away immediately after birth (Lewis, 1976; Lasker & Toedter, 1994). This approach received considerable criticism however, and policies were changed to allow parents to have contact and make memories with their stillborn baby (Lasker & Toedter, 1994; Cunningham, 2012). Qualitative studies found this to be a positive change, with parents expressing that the opportunity to see and hold their baby was invaluable (Gold, Dalton & Schwenk, 2007; Downe et al., 2013; Mills et al., 2014). Despite support for such practices in the qualitative literature, sparse quantitative data exist on the impact of contact with the stillborn, and mixed results have been noted in studies that have been conducted (Koopmans, Wilson, Cacciatore & Flenady, 2013). Inconsistency in the quantitative literature has led to confusion regarding the best evidence-based care for parents. Clinical guidelines have differed in the recommendations provided, with NICE guidelines receiving considerable criticism for including recommendations not to encourage parents to see or hold their stillborn (NICE, 2006; 2007; Cunningham, 2012; Downe et al., 2013; Koopmans et al., 2013; Cacciatore, 2013; Ryninks, Roberts-Collins, McKenzie-Mcharg & Horsch, 2014).

The present work will provide a systematic review of quantitative studies investigating the impact of contact with the stillborn after birth on mothers’ and partners’ wellbeing. Extant evidence for the benefits and harms will be collated and studies appraised for risk of bias and quality. Further, this review will seek to highlight moderating factors, drawn from the literature, which may influence the relationship between contact with the infant and outcomes (e.g., care providers’ support, gestation of stillbirth, subsequent pregnancy) (Erlandsson, Warland, Cacciatore & Radestad, 2013; Radestad et al., 2009; Cacciatore, Radestad & Froen, 2008).
Objectives

The purpose of this review is to collate and critically appraise extant evidence for the impact of contact with the stillborn infant on mothers’ and partners’ mental health and wellbeing.

Methods

This protocol has also been registered on PROSPERO (CRD42014013890) and can be accessed at: http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42014013890#.VCQavRY09p0

Criteria for considering studies for this review

Types of studies

We will include all studies which quantitatively compare the outcomes for women and partners who held or did not hold their stillborn baby. It is not expected that any randomised controlled trials could be ethically conducted. Prospective and retrospective cohort and case-control studies, and cross-sectional studies will be eligible for inclusion. Qualitative studies which do not provide any quantitative comparison of outcomes will not be included.

Types of participants

We will include studies of all women and partners who had a singleton or multiple stillbirth. Studies will not be excluded based on the gestational definition of stillbirth, as this varies between jurisdictions. However, the gestation at stillbirth and gestational definition of stillbirth used in included studies will be noted.

Types of contact

The primary intervention of interest is whether women/partners held their stillborn baby in the hours or days after birth. Rates of seeing the stillborn infant will be recorded in data extraction, however, are anticipated to be near universal so will not be considered an intervention for comparison. Secondary interventions of interest include other interaction with the stillborn or memory making activities which parents may undertake in recognising the presence of their baby and in building memories, for example, bathing the baby, dressing the baby, taking photos, and taking hand/footprints.

Outcome measures

Primary outcomes

1. Clinically diagnosed mental health problems (e.g., depression, anxiety, post-traumatic stress disorder)
2. Standardised assessment of mental health issues (e.g., Edinburgh Postnatal Depression Scale, Beck Depression Inventory, Depression Anxiety Stress Scales)
3. Self-reported poor mental health or symptoms of psychological distress (non-standardised/validated surveys)

**Secondary outcomes**

1. Self-reported measures of poor maternal/partner health identified by stakeholders (Redshaw, Rowe & Henderson, 2014):
   a. Poor physical health
   b. Fatigue or severe tiredness
   c. Sleep problems
2. Personal relationship difficulties
3. Satisfaction with decision/opportunity to hold or not hold their stillborn baby

**Search methods for identification of studies**

The search strategy was developed based on inclusion criteria. In addition, searching strategy reported in a recent qualitative synthesis (Mills et al., 2014) was consulted, as was the *Cochrane Pregnancy and Childbirth Group* specialised register MeSH terms. A search technician provided guidance on the searching strategy.

Searches will be conducted in English, but no language restrictions will be set. No restrictions will be set by date or publication type.

**Electronic searches**

The following databases will be searched from inception to present:

- Applied Social Science Index and Abstracts (ASSIA)
- British Nursing Index (BNI)
- Cochrane Database of Systematic Reviews
- Cochrane Pregnancy and Childbirth Group Trial Register
- Cumulative Index to Nursing and Allied Health (CINAHL) plus
- EMBASE
- Health Services Research Projects in Progress (HSRProj)
- Medline
- Open Grey
- PsycINFO
- ProQuest Dissertations and Theses
- Science Citation Index
- Social Sciences Citation Index
Search strategy for Medline is displayed in Box 1 and will be adapted for other databases accordingly.

**Box 1. Searching strategy for Medline**

<table>
<thead>
<tr>
<th>Search #1:</th>
<th>Exp stillbirth/ OR exp fetal death/ OR abortion, spontaneous/ OR perinatal mortality/ OR (&quot;fetus death&quot; OR &quot;fetus loss&quot; OR &quot;foetal loss&quot; OR &quot;foetal death&quot; OR &quot;fetal death&quot; OR &quot;fetal loss&quot; OR &quot;neonat* death&quot; OR &quot;neonatal loss&quot; OR &quot;newborn death&quot; OR &quot;newborn loss&quot; OR &quot;perinatal death&quot; OR &quot;perinatal loss&quot; OR &quot;pregnanc* loss&quot; OR &quot;stillb*&quot; OR &quot;still born&quot; OR &quot;still birth&quot;).mp.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Search #2:</td>
<td>Exp maternal behavior/ OR paternal behavior/ OR touch/ OR rooming-in care/ OR (contact OR held OR hold* OR touch* OR bath OR bathing OR &quot;care practice*&quot; OR &quot;care guideline*&quot; OR footprint* OR handprint* OR memory OR memories OR momento OR photograph* OR policy OR policies OR &quot;psychosocial care&quot; OR wash OR washing).mp.</td>
</tr>
<tr>
<td>Search #3:</td>
<td>Exp adaptation, psychological/ OR exp anxiety/ OR exp anxiety disorders/ OR Exp depression/ OR exp depression postpartum/ OR depressive disorder/ OR Exp mood disorders/ OR exp grief/ OR Exp mental health/ OR mental disorders/ OR Adjustment Disorders/ OR exp stress disorders, post-traumatic/ OR stress, psychological/ OR sleep disorders/ OR &quot;sleep initiation and maintenance disorders&quot;/ OR (adjustment OR anx* OR coping OR depress* OR distress OR divorce OR fatigue OR &quot;interpersonal difficult*&quot; OR &quot;interpersonal problem*&quot; OR &quot;interpersonal trouble*&quot; OR &quot;insomnia&quot; OR &quot;mental health&quot; OR &quot;mental disorder&quot; OR &quot;mental illness&quot; OR &quot;physical health&quot; OR &quot;poor sleep&quot; OR &quot;posttraumatic stress&quot; OR &quot;post-traumatic stress&quot; OR &quot;postnatal anxiety&quot; OR &quot;postnatal depression&quot; OR &quot;postpartum anxiety&quot; OR &quot;postpartum depression&quot; OR &quot;puerperal depression&quot; OR &quot;puerperal stress&quot; OR psychological OR psychosocial OR PTSD OR regret* OR &quot;relationship break*&quot; OR &quot;relationship difficulty*&quot; OR &quot;relationship dissolution*&quot; OR &quot;relationship problem*&quot; OR &quot;relationship trouble*&quot; OR satisfaction OR satisfied OR stress* OR &quot;sleep problem*&quot; OR &quot;sleep difficult*&quot; OR &quot;sleeping problem*&quot; OR &quot;sleeping difficult*&quot; OR tired* OR wellbeing OR &quot;well being&quot; OR &quot;well being&quot; OR &quot;well-being&quot;).mp.</td>
</tr>
<tr>
<td>Search #4:</td>
<td>Search #1 AND Search #2 AND Search #3</td>
</tr>
</tbody>
</table>

**Searching other resources**

1. Additional grey literature will be sought by searching the websites of the Stillbirth and Neonatal Death Charity (SANDS) and International Stillbirth Alliance (ISA)

2. We will search the reference lists of all studies meeting inclusion criteria, and forward citations of studies meeting inclusion criteria.

3. We will contact subject experts to identify unpublished or ongoing research

**Data collection and analysis**

**Selection of studies**

Titles and abstracts returned from searches will be screened independently by two reviewers (JMH and JH) and appraised in light of inclusion criteria. Where reviewers independently determine studies do not meet inclusion criteria, they will be excluded. Full records will be obtained for studies meeting inclusion criteria or with insufficient information to assess inclusion criteria from the title and abstract. Two reviewers (JMH and JH) will independently screen full-text articles and assess consistency with inclusion criteria. Where there is disagreement, reviewers will meet and seek to reach consensus, and studies will be referred to a third review author (MR). Full selection process for study screening and inclusion will be recorded in accordance with current guidelines (Moher, Liberati, Tetzlaff & Altman, 2009).
Data extraction and management

Data will be extracted from papers using a piloted data extraction form developed for this review. Two reviewers (JMH & JH) will independently extract data from each included study, where disagreement occurs judgement will be referred to a third review author (MR).

Data extraction will include study details, methods, participants (e.g., response rate, sample representativeness, parity, age, gestation of stillbirth, % currently pregnant), intervention details (e.g., assessment of contact with the stillborn, data on duration or nature of contact with stillborn, staff behaviour), outcomes (e.g., timing of outcome assessment, confounds included in adjustments, mode of data collection), subgroup details (e.g., subgroups included in study and differences identified by subgroups), funding, author contact details and study quality assessment (see below).

Quality assessment of included studies

Whilst there are clear guidelines and evidence for the assessment of risk of bias in randomised controlled trials (see Higgins & Green, 2011), risk of bias assessment for observational research has yet to be agreed upon and remains controversial (Sanderson, Tatt & Higgins, 2007). The risk of bias and quality of included studies will be assessed for this review using a checklist developed based on the items of the Strengthening the Reporting of Observational studies in Epidemiology (STROBE) statement (von Elm et al., 2007), Critical Appraisal Skills Program (2013) checklists for cohort and case-control trials, and the Newcastle-Ottawa Scales (NOS) for assessing the quality of nonrandomised studies (Wells et al., 2004), based on recent appraisal of quality assessment tools for observational studies (Sanderson, Tatt & Higgins, 2007).

The following items will be appraised for each study and rated as high risk, low risk or unclear risk:

- Sample representativeness (reflected through adequate recruitment, exclusion criteria, response rates, and comparability to wider birthing or stillbirth population)
- Adequacy of exposure measurement (adequacy of the assessment of contact with the stillborn infant)
- Incomplete outcome data (attrition bias)
- Selective outcome reporting
- Other bias
- Two additional items will be appraised capturing group comparability and statistical adjustment for potential confounds:
  - Comparability of exposed and non-exposed participants
  - Adequacy of statistical methods and confound adjustment
These two items will be rated as high risk (little comparability/no adjustment), comparable/adjusted on demographic characteristics, or comparable/adjusted on more specific characteristics for these comparisons (e.g., pre-pregnancy mental health issues, mode of delivery), representing the lowest risk of bias. This rating will be used to provide more nuanced appraisal of study quality and is consistent with recent evidence that the results of non-randomised designs more closely approximate those of randomised studies when adjustment is made for relevant area-specific characteristics, rather than characteristics of convenience such as demographics (Shadish, Clark & Steiner, 2008).

For each study, two reviewers (JMH & JH) will independently appraise quality. Where reviewers cannot reach consensus regarding the categorisation of bias for a particular study, judgement will be referred to a third review author (MR).

Data synthesis

We anticipate that only a small number of heterogeneous studies will be eligible for inclusion in the review. We do not anticipate that quantitative synthesis will be possible. Thus, narrative synthesis of studies will be provided, consistent with current guidelines (Moher et al., 2009; Higgins & Green, 2011). Measures of effect for each study will be presented. Odds and risk ratios for dichotomous outcomes and mean differences or standardised mean differences for continuous outcomes will be calculated to represent the aggregate difference in outcomes between those who held or did not hold their stillborn baby (or engaged in other memory making activities). Where adequate adjustment has occurred for potential confounds, adjusted measures of effect will be provided in preference to raw comparisons. Where outcomes are reported for multiple time points, data will be extracted and presented for each time point. In addition, measures of effect for key subgroups/proposed moderators (see below) will be presented. If statistics are missing they will be calculated from other data provided (where possible). Where missing statistics cannot be calculated, authors will be contacted for further information. Where authors cannot be reached or do not provide a response, data will be listed as missing.

Sub-group comparisons

From review of background literature the following factors are anticipated to influence the association between contact with the stillborn baby and mental health and wellbeing outcomes:

1. Time since the stillbirth (timing of maternal/paternal outcome assessment)
2. Women pregnant at the time of outcome assessment
3. Subsequent live birth/s
4. Gestation of stillbirth
5. Time from antepartum death to birth
6. Level of support for contact/memory making provided by staff

Where possible outcomes will be stratified according to these sub-groups/conditions and changes to any relationship between contact/memory making and outcomes will be noted.
References


