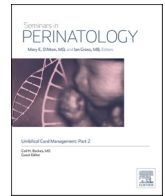


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Care in pregnancy after stillbirth

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ARTICLE INFO

Keywords:

Pregnancy after loss
Pregnancy after stillbirth
Psychological support

ABSTRACT

Pregnancy after stillbirth is associated with increased risk of stillbirth and other adverse pregnancy outcomes including fetal growth restriction, preeclampsia, and preterm birth in subsequent pregnancies. In addition, pregnancy after stillbirth is associated with emotional and psychological challenges for women and their families. This manuscript summarizes information available to guide clinicians for how to manage a pregnancy after stillbirth by appreciating the nature of the increased risk in future pregnancies, and that these are not affected by interpregnancy interval. Qualitative studies have identified clinician behaviors that women find helpful during subsequent pregnancies after loss which can be implemented into practice. The role of peer support and need for professional input from the antenatal period through to after the birth of a live baby is discussed. Finally, areas for research are highlighted to develop care further for this group of women at increased risk of medical and psychological complications.

Introduction

This article addresses pregnancy after stillbirth; we have chosen this topic as an area of focus due to the increased incidence of fetal, maternal, and neonatal complications and psychological morbidity experienced in pregnancy after loss. Approximately 60 % of women who experience a stillbirth go on to have a subsequent pregnancy.^{1,2} In some cases this may be soon after the loss of their baby; a survey of 275 women in the USA found a median interpregnancy interval of 6 months (interquartile range (IQR) 4-10 months).³ Pregnancies after stillbirth are associated with greater resource use, including increased rates of induction of labor (IOL) and of Cesarean delivery than women with a history of live birth.^{1,4} Thus, it is important that women receive optimal care in pregnancy after loss to minimize the risk of subsequent complications and to avoid excessive intervention. This article will consider pregnancy after stillbirth by identifying evidence to answer key questions.

What are the medical consequences of pregnancy after stillbirth?

Understanding the relationship between a previous stillbirth and the

risk of adverse outcome relies on two types of studies. Epidemiological studies (and systematic reviews thereof) have demonstrated that women with a prior history of stillbirth have increased risk of stillbirth and other related adverse pregnancy outcomes. However, due to a lack of information about individual cases these studies cannot determine whether this relationship is mediated by specific conditions. A series of smaller studies have identified individual maternal characteristics and causes of stillbirth which are associated with adverse outcomes in a subsequent pregnancy.

A large systematic review including 3,412,079 women from 13 cohort and 3 case-control studies found a stillbirth rate of 2.5 % in women with a history of stillbirth compared to 0.4 % in women with a history of live birth (pooled Odds Ratio (OR) 4.83 95 % Confidence Interval (95 % CI) 3.77, 6.18); when this was adjusted for potential confounding factors the pooled OR was 3.38 (95 % CI 2.61, 4.38).⁵ A study using the Swedish national maternity dataset found that the risk of adverse outcome persisted in subsequent pregnancies (i.e. a third pregnancy when the first ended in stillbirth, and the second ended in live birth, adjusted OR (aOR) 2.35, 95 % CI 1.68, 3.28).⁶ This risk was independent of the gestation of stillbirth and the presence of maternal medical disorders related to stillbirth (diabetes and hypertension).

Supported in part by Tommy's Charity and Manchester University NHS Foundation Trust. Disclosure: The authors report no potential conflicts of interest. (Authors please confirm complete and correct)

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<https://doi.org/10.1016/j.semperi.2023.151872>

Available online 20 December 2023

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A history of stillbirth also increases the risk of maternal and neonatal complications in subsequent pregnancies. In their study of 364 women who had a stillbirth in the Grampian region of Scotland, women who had a stillbirth in the first pregnancy had an increased risk of preeclampsia, placental abruption, and preterm birth. In addition, these women had an increased incidence of obstetric intervention including induction of labor (IOL) and Cesarean delivery.⁷ These increased rates of intervention were confirmed by a further study from Ireland which found IOL in 48.1 % and Cesarean birth in 40.5 % of women;⁴ the history of stillbirth was cited as the indication for IOL in 63.5 % of cases indicating that intervention may be driven solely by the history of stillbirth rather than the presence of recent pathology. The cohort study using Swedish national maternity data demonstrated that the risk of complications, specifically: preterm birth, preeclampsia, and placental abruption also persisted in pregnancies following stillbirth, not just the immediate pregnancy after loss.⁶ However, it is important to appreciate whether the cause of stillbirth or other maternal factors have an impact on the recurrence of stillbirth.

Nijkamp et al. explored 163 perinatal deaths (over 16 weeks' gestation) in the Netherlands. They found recurrent fetal death in 11 cases (6.7 %), in which the cause was found in 7 cases, including early preterm prelabor rupture of membranes and placental-bed pathology.⁸ An Italian study of 273 pregnancies after stillbirth found adverse outcome in 24.5 %. This study found an adverse outcome was more common in women who had placental vascular disorders compared to those who had an unexplained stillbirth and also women who had maternal obesity.⁹ This study did not find a relationship between cigarette smoking and subsequent adverse outcome (OR 1.9, 95 % CI 0.9, 4.7) though this could be due to small numbers of women who smoked cigarettes (9.2 %).⁹ A study of 266 pregnancies from the UK described adverse outcome in 69 cases (25.9 %) including three further perinatal deaths.¹⁰ Women were more likely to have an adverse outcome if there was a pre-existing medical condition (aOR 2.12, 95 % CI 1.10, 4.12) and they continued to smoke cigarettes (aOR 6.80, 95 % CI 1.99, 23.30).¹⁰ This study did not find a relationship between the gestational age of stillbirth or classification of the cause of stillbirth and subsequent outcome. Abnormalities of the placenta including: maternal vascular malperfusion, fetal vascular malperfusion, and chorioamnionitis were associated with increased risk of adverse outcome in the subsequent pregnancy. A study of 128 women who had histological evidence of placental infarction (consistent with maternal vascular malperfusion) found 31 % had adverse outcomes.¹¹ These studies suggest that using data regarding maternal characteristics and the cause of the index stillbirth could provide valuable information to guide management of subsequent pregnancies.

What is the effect of interpregnancy interval?

Patients report wide variation in the recommended interpregnancy interval (IPI) with a median of 6 months (IQR 2-9); the median recommended IPI was greater if the previous birth was by Cesarean delivery (9 months vs. 4.2 months), but there was no difference in recommended IPI if women were older.³ The impact of IPI has been evaluated in several large studies. While the risk of perinatal death is increased for IPI less than six months after a live birth in the wider population, this effect was not seen in pregnancies following miscarriage or stillbirth.¹² A large international cohort study using records from Finland, Norway, and Western Australian found in 14,452 women who had a stillbirth, the median IPI was 9 months (range 4-19 months). As noted previously, women with a history of stillbirth in this cohort had a high risk of stillbirth (2 %), preterm birth (18 %), and small for gestational age (9 %). There was no association between any of the IPIs and the risk of subsequent stillbirth, preterm birth, or adverse outcome.¹³ Another recent cohort study of 5,581 women in the USA found that none of the IPI categories was associated with increased risk of adverse pregnancy outcome. This absence of a relationship between IPI and outcome after

pregnancies ending in stillbirth may reflect the higher risk of adverse outcome in pregnancy after stillbirth, which diminishes the relative effect of IPI. It should be recognized that when counselling parents about appropriate time to try to conceive that both partners may have differing psychological or emotional drives regarding timing of a future pregnancy. However, parents can be reassured that the duration of IPI does not alter the likelihood of subsequent adverse outcome.

What are optimal care models for pregnancy after stillbirth?

Current clinical practice guidelines provide some outline recommendations for care of pregnancy after stillbirth (though these are acknowledged to be based on low-grade evidence). The Royal College of Obstetricians and Gynecologists (RCOG) states that i) parents should be advised about the cause of stillbirth, the chance of recurrence, and any specific means to prevent further loss; ii) women should be offered general pre-pregnancy advice including support for smoking cessation; iii) women should be advised to avoid weight gain if they are already overweight and consider weight loss and iv) an offer should be made to discuss the potential benefit of delaying conception until severe psychological issues have been resolved.¹⁴ As women with a prior stillbirth are also at increased risk of having a small for gestational age infant, the RCOG recommend that they should have serial ultrasound measurement of fetal size and assessment of fetal wellbeing with umbilical artery Doppler from 26-28 weeks of pregnancy.¹⁵

An international consensus statement for the management of pregnancy after stillbirth was published in 2018; this was developed by a multidisciplinary group of 27 professionals and parents with lived experience.¹⁶ This adds to existing guidance by noting that low-dose aspirin may reduce the risk of perinatal death in women at risk for placental insufficiency (including women with a stillbirth from placental causes). The consensus statement notes that decisions around timing of birth should incorporate the circumstances surrounding the previous stillbirth, the clinical picture of the current pregnancy, and the emotional state of the mother and her family, while balancing these against the negative effects of birth prior to 39 weeks. Thus, there may be a role for early term (37–39 weeks) birth, but there is no evidence for delivery before 37 weeks' gestation based on a history of stillbirth alone. Further recommendations are made, largely focussing on the need for emotional support from professionals and peers. Critically, care provision is recommended to include consistent and timely medical and psychosocial care, services, and support by skilled and familiar care teams knowledgeable about the pervasive impact of stillbirth on the subsequent pregnancy and beyond.

Models describing optimal care in pregnancy after stillbirth largely use information about "what works" from surveys or qualitative studies of pregnancy after loss. A summary of approaches and behaviors associated with positive experiences of pregnancy after loss (PAL) services are shown in [Table 1](#); these have been grouped into three areas relating to the delivery of specialist care: sensitive communication and conduct of staff, organisation of services, and maternal and fetal monitoring and surveillance. It is important that these features are not seen as a "to do" list, but one that allows elements to be individualized to each woman and her family as "ideal" care varied between study participants.

Women and families have shared their experiences of pregnancy after stillbirth to provide evidence on their preferred elements of respectful, skilled, and compassionate care. Unsurprisingly, descriptions of negative care experiences are often the opposite of what families report as supportive and important in the care they receive in pregnancy after stillbirth. Sadly, many families still report care that does not meet their needs or expectations. An international survey of 2,716 parents found that only around half of all respondents felt that elements of quality, respectful care were consistently applied, care varied widely, and care addressing psychosocial needs was less frequently provided than additional medical interventions.² Similarly, a UK cross-sectional survey completed by 547 women reported both positive and negative

Table 1
Summary of health professional's behaviors and approaches identified as relating to positive service user experience in pregnancy after stillbirth.

Area	Approach / behavior	References
Sensitive communication and conduct	Engagement with other family members, partner, living children etc.	27, 28
	Acknowledgment and discussion of previous loss	17, 19, 25-29
	Use of previous baby's name	29
	Emotionally supportive relationship with carers	17, 19, 25-28, 30, 33, 36
	Validation – knowledge that similar feelings were experienced by other parents with a history of previous pregnancy loss	17, 19, 26-30
	Organisation of services	Easy to access by various means (call/text/email)
Monitoring and surveillance	Continuity of carer (obstetrician and midwife)	17, 19, 26-30, 33, 36
	Flexibility of appointments	17, 19, 26, 28
	Provision of information	17, 19, 26-30
	Additional appointments	17-19, 26, 28, 29, 30
	Ability to avoid contact with pregnant women with no history of loss	19
	Access to peer support	17-19, 26-29
	Increased visits for cardiotocography / non-stress test or blood pressure tests	17, 19, 26, 28-30
	Increased access to ultrasound scans	17, 19, 26, 28-30

experiences with the healthcare system and professionals, mainly around interactions with staff and the organization and delivery of services.¹⁷ Women negatively remembered professionals who were not aware of their history despite it being shared with the healthcare team or in their chart, who minimized or misunderstood their thoughts and feelings, and who expressed doubts about the need for additional medical surveillance, interpreted as a lack of knowledge about the pervasive impact of stillbirth.¹⁷ Women also reported feelings of isolation^{18,19} and experience higher rates of anxiety and depression in pregnancies after stillbirth compared to women with a history of a live birth, or no births.^{18,20-24}

Six themes were identified from 62 families from Canada regarding their experience of pregnancy after stillbirth and their recommendations for improving care. These were: desiring recognition and acknowledgment of anxiety, parental voices to be heard and taken seriously, the addition of specific clinical care for reassurance, kindness and empathy, peer support, and an understanding of the general experience including guilt, self-blame, the balance between positive and realistic thoughts, and challenges with a lack of continuity of care providers.²⁵

In addition, an Australian study found that improved social connectedness during a pregnancy after stillbirth decreased feelings of self-blame and despair, and that midwives were well-positioned to provide supportive relationships to foster these connections.²⁶ This is corroborated by another Canadian study of 33 women that found that social support from both professionals and peers helped to foster hope in subsequent pregnancies, and that women continued to balance hope with fear in the context of changing information and past experiences.²⁷ The authors concluded that professionals can make a positive impact on the care experience with meaningful actions such as the provision of information, acknowledgment of grief, screening and connection to early and ongoing supports, and by spending time to listen and to assess changing needs.²⁷ In an evaluation of an Australian pregnancy after loss clinic, 10 women positively rated being followed by an experienced multi-professional team known to them, especially in having recognition and validation from professionals that they may experience mixed or challenging emotions such as fear, anger, sadness, guilt, and hope.²⁸ Families also highly valued peer support, flexible appointments, advocacy, and emotionally supportive relationships with professionals that

were responsive to their changing needs and able to anticipate and normalize potential experiences.²⁸ While both additional medical and emotional care were important, families most valued the emotional care they received.²⁸ Similarly, qualitative interviews of 20 women attending a specialist antenatal service in the UK for pregnancies after perinatal death found women appreciated expert caregivers who knew their history, listened to their concerns, and provided realistic information and support.¹⁹ Women reported balancing hope for the pregnancy with personal risk appraisals; supportive relationships with clinic professionals helped to ease stress and foster feelings of hope.¹⁹ The use of sensitive language and provision of information to minimize uncertainty is important to families.^{29,30}

Professionals who listen to family feedback and implement personalized care have the potential to profoundly alter the care experience. Families consistently indicate that any professional who comes into contact with them during their pregnancy care has the opportunity to make both a positive or negative difference with the care they provide.^{2,17,29-31} Recent reports on specialist clinics and suggestions for potential comprehensive care models^{2,16,19,28,31} highlight the importance of skilled multi-professional teams with the ability to individualize care, who also know the families and their histories and care preferences. Parents have described the positive impact of primary care providers (physicians, midwives, and nurses)^{21,26,28,29} and allied health professionals (mental health and public health professionals, sonographers, social workers)^{18,21,27-29,32} on their experience of supportive and compassionate care, as well as many interactions that fall short of parental needs or expectations.^{2,17,27-30} Consistently, the provision of information such as recurrence risk and strategies to reduce risk^{29,30}, support and maintenance of hope without minimizing valid fears,^{27,29,30} early and ongoing connection to supports, including peer supports^{18,28,29}, additional monitoring and surveillance^{17,29-31}, joint decision-making^{2,19,33}, and compassionate listening were all highly valued by families.^{4,17,27,28,30}

To date there have been no large-scale comparative studies of the care provided by PAL services and the effect on maternal and neonatal outcomes. A feasibility study of 38 women offered a study intervention (of continuity of midwife care and access to group and online support) found those who received increased midwifery continuity valued the relationship with the care coordinator and perceived positive impacts on pregnancy experiences. A retrospective review of women attending a single centre service found a reduced stillbirth rate (2/94 before vs. 0/84 after the introduction of PAL service) and lower preterm birth rates (20/94 before vs 8/84 after introduction, $p=0.03$).³⁴ A large-scale analysis of the impact of PAL services over time is underway in the UK.

What is the role for peer support in pregnancy after loss?

There is a paucity of research exploring or evaluating different models of peer support in pregnancy after loss. Westby et al. (2021) found that social support, amongst other factors, affected levels of symptoms for anxiety, depression, and PTSD.³⁵ Social support (along with high quality clinical care following history of stillbirth) is identified as being an important factor in promoting ongoing parental mental health and wellbeing; peer support is proposed to have a similar impact. In their online survey, Gower et al. found parents with experience of PAL frequently sought support options that involved connection to others with similar experiences.²⁵ How this might be supported (e.g. by signposting to existing local support groups or facilitating PAL clinic-specific groups) and what role should be adopted by healthcare professionals, is worthy of research.

Mills et al. evaluated peer support (as a component of a care package also including care and contact with a named midwife coordinator).³⁶ This was a small feasibility study of 38 women (across pre- and post-intervention phases) in two UK maternity units. Clinical and psychological outcomes were measured as well as qualitative interviews conducted to develop insight into experiences of research involvement

and care components. The study retention rate was good (more than 75 %), but the in-person support group was only accessed by 5 (13 %) of women. The online group was accessed by 21 (55 %) of women, but engagement with this decreased over time.³⁶ Interviews revealed that the intervention (care package) was viewed as a good idea but implemented variably. Importantly, the study intervention involved specific training for midwife care-coordinators who were assigned no more than three women to avoid overwhelming the service, but the study suffered from care coordinators being unable to maintain the level of contact they would have liked (challenges revealed in interviews included service pressures, roster changes, annual leave, and part time working schedules). Nevertheless, relational care and means of supporting effective peer support in pregnancy after loss remain worthy of further exploration.

Peer support in pregnancy after loss appears to be a nuanced and delicate matter, challenged by the likely diverse or vacillating profile of behaviors and coping strategies used by individual women and within any given cohort of women pregnant after loss. Women move between loss-orientated and hope-orientated thoughts and actions and it seems likely that this would make regularly engaging with a strict format of group meetings difficult. Smith et al. report that women pregnant after loss avoid other women who are pregnant without the same loss history as they feel guilty about exposing them to realities around stillbirth risk.¹⁹ This can isolate women and often women are reluctant to attend general antenatal classes. Thus, peer support seems relevant and potentially beneficial as a supportive mechanism. However, how peer support can be facilitated practically and sensitively needs further investigation.

Care provision after the birth of a live baby

There is very little research exploring or evaluating experiences of care, or needs, in the postnatal period following pregnancy after stillbirth. The emotional and psychological challenges faced by women in the antenatal period have significant potential to continue into the postnatal period. These issues are likely to include: ongoing anxiety, depression, trauma, grief, social isolation, and challenges with relationships and adapting to parenthood. A previous stillbirth is an established risk factor for postnatal depression.³⁷ The postnatal period may also trigger fresh grief around what was lost or not experienced with the baby that died. Additionally, in the postnatal period the partner roles change to become co-parents, and it is established their experiences and needs are under-researched.

In a longitudinal study of 204 mothers Côté-Arsenault (2020) found a relationship between the gestation of prior loss and mothers' feelings about parenting and attachment style of the child born subsequently.³⁸ Losses at later gestations were associated with more negative feelings and less secure attachment. Caldwell et al. suggest interplay between factors related to adult attachment, shame, and grief and coping, with social connectedness appearing to be a protective factor.²⁶ Although this study did not focus on the postnatal period, the authors recommended that clinicians focus on social connectedness as a means of supporting families navigating PAL. There is an opportunity to sow the seeds antenatally to enable women to redevelop social networks that are often placed on hold during an anxious pregnancy.

Studies of specialist PAL services provide further evidence of the need for postnatal support. Women who had been cared for in a specialist PAL clinic valued this service but felt that specialist postnatal care was disproportionately lacking.¹⁹ Meredith et al. found women in PAL clinic frequently sought to continue contact with their antenatal midwife in the postnatal period.²⁸ The authors recommended a "formal termination" of the therapeutic relationship which should be embedded in care planning. Anecdotally, events when families can meet the clinical team informally (e.g. annual parties or events) are valued and offer closure of that episode of care. It is important that the evidence of unmet needs in the postnatal period of PAL are incorporated into future studies.

What are the gaps and future research opportunities?

As noted above, comprehensive evidence-based guidelines for the management of pregnancy after stillbirth are lacking, and wide variations in care are reported by families and professionals in both care after the index stillbirth and into subsequent pregnancies.^{2,17,28} Many professionals do not have a way to receive feedback, either formal or informal, on the care they provide and may believe they are meeting the needs of families. The most effective and efficient ways to implement awareness, education, and training remain unknown.^{29,30} Finally, while several interventions have been reported to be supportive for families,^{19,26,28,31,33,39} it remains to be seen if these interventions or care models are feasible in diverse healthcare settings³⁶ and effective and acceptable to a broader range of families, especially those from diverse cultural and psychosocial backgrounds.^{26,27,39}

Identifying optimal care for pregnancies following stillbirth was identified as one the top ten research priorities by the UK Stillbirth Priority Setting Partnership conducted in 2014.⁴⁰ A subsequent research prioritization exercise including 79 individuals involved in clinical practice, parent advocacy, and stillbirth research ranked 16 areas for priority. The top seven areas for research in subsequent pregnancy were all ranked as important and urgent by > 70 % of respondents, these were: medical therapies for placental dysfunction (81 %), additional antepartum fetal surveillance (80 %), core outcomes dataset for stillbirth research (79 %), targeted antenatal interventions (79 %), risk level by cause of stillbirth (79 %), experience and care in low and middle income countries (75 %), and specialist clinical services (73 %).⁴¹ Thirty-eight additional topics were suggested including education and support for healthcare providers, and further suggestions regarding psychosocial care of women and their families.

The means by which research evaluation can be conducted was also considered by participants. The most robust means of evaluation e.g. randomized controlled trials (RCTs), was reported to be the "best way to evaluate interventions" by 39 % (different forms of psychological support) to 72 % of respondents to the priority setting exercise (medical therapies for placental dysfunction) of responses.⁴¹ Whilst it was felt by a high proportion of respondents that RCT-grade evidence would improve implementation, fewer than 50 % of respondents felt that this would be feasible. This was driven to some extent by concerns that RCTs would need to be large to demonstrate difference in outcome. It is worth noting that the systematic review examining interventions to improve outcome in pregnancies after stillbirth included only 10 RCTs with 222 participants.⁴² Therefore, it is likely that trials to address these prioritized questions would need to be international collaborations to achieve adequately powered studies.

There is ongoing need to evaluate the psychological impact of a subsequent pregnancy into the postnatal period and its longer-term effects on maternal-fetal bonding. While there is some preliminary evidence, additional studies to describe parents' views and experiences about their journey of pregnancy after loss are needed to identify and describe good practice. This information can then be used to inform models of care and clinical trials of these models.

While it is important to recognize areas for future research, it is also important to acknowledge that significant improvement in current care practices could immediately be seen if professionals reflected on gaps in providing compassionate and skilled care to families and prioritized listening to what families have already shared, especially as related to psychosocial interventions, which are less likely to be included in current care models, but very likely to benefit the family.^{19,27,28,43}

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