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Anxiety and acceptability related to participation in stillbirth research



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ARTICLE INFO

Article history:

Received 2 February 2015

Received in revised form

16 July 2015

Accepted 16 July 2015

Keywords:

Stillbirth

Anxiety

Satisfaction

Bereavement

Research participation

ABSTRACT

Background: stillbirth research is often hampered by the need to 'protect' both bereaved families as well as healthy pregnant women from distress resulting from recruitment by research staff. No studies have investigated anxiety levels of recently bereaved or healthy pregnant women participating in stillbirth research. The aim of this study was to assess anxiety levels and acceptability of women participating in a stillbirth case-control study.

Method: a follow-up questionnaire was posted to all participants of the Sydney Stillbirth Study in 2012. The questionnaire assessed the anxiety level experienced by women as a result of their participation in the study. Questions related to the initial approach of the research staff; level of anxiety at time of consent and after the interview; and reasons for and satisfaction with participation. The Spielberger (STAI-6) anxiety scale and open-field responses were included.

Results: 35/103 case participants and 65/192 control participants returned the completed questionnaire. The majority participated for altruistic reasons. 20/35 (cases) and 58/65 (controls) stated they disagreed/strongly disagreed that participation in the study increased their anxiety. 1 in 5 cases reported that participation in the study increased their anxiety; however this did not affect their satisfaction. Timing of interview did not affect anxiety scale responses. ($F=1.2$; $p=0.37$) 30/35 (cases) and 63/65 (controls) stated they agreed/strongly agreed that they were satisfied participating in the study.

Conclusions: these findings suggest high levels of satisfaction amongst both case and control participants and no statistically significant increase in anxiety related to involvement in stillbirth research. 'Protecting' families may require further justification.

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Background

Stillbirth is one of the most devastating losses a parent can experience, and is associated with an increased risk of long term anxiety related symptoms (LaRoche et al., 1984; Radestad et al., 1996; Kelly and Trinidad, 2012; Cacciato, 2013). Stillbirth research is often hampered by 'gatekeepers' such as ethics committees, hospital staff and even close family members who try to protect bereaved families from undue distress potentially resulting from recruitment by research staff at such a vulnerable time (Dent et al., 1996; Sque, 2000; Dyregrov, 2004; Buckle et al., 2010). Healthy, pregnant women

are also perceived to be in a 'vulnerable' condition and are subject to the same sort of well-meaning protective behaviour (Wild, 2012). There is general uncertainty regarding recruitment methods and timing, and concern about intensifying distress or anxiety that does not result in direct benefit to the participants (Scott et al., 2002; Burnell and O'Keefe, 2004; Kreicbergs et al., 2004).

In our experience of conducting a case-control study into stillbirth (the Sydney Stillbirth Study) (Gordon et al., 2015), we found that two of the greatest barriers to obtaining ethics approval and overcoming staff resistance in recruiting potential study participants related to (1) inviting women to participate in research soon after receiving the news that their baby had died and (2) 'cold-calling' healthy pregnant women to participate in such a sensitive area of research.

These ethical issues are a valid concern, particularly in light of the growing global interest in stillbirth. Despite this, there is little empirical work examining the responses of bereaved individuals to participation in research (Beck and Konnert, 2007). The studies that

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have done so include bereaved participants who had experienced the loss of a parent, spouse or other family member (Cook, 1995; Buckle et al., 2010), the loss of a child or other family member due to cancer (Seamark et al., 2000; Scott et al., 2002; Kreicbergs et al., 2004), parents who lost their child through Sudden Infant Death Syndrome, suicides or accidents (Dyregrov, 2004), and parents who lost their child through a chronic progressive condition (Hynson et al., 2006). One study that recruited via on-line grief support groups examined the theoretical opinions of bereaved adults regarding ethical issues in bereavement research, however only six of the 316 respondents had previously participated in a research project (Beck and Konnert, 2007). The time since the loss in these studies varied from less than one month to nearly 10 years. The only study which included parents of stillborn babies were interviewed between three and just over nine years after the loss (Brabin and Berah, 1995). Contrary to the concerns commonly expressed about bereavement research, all of these studies indicate a positive response by the majority of participants as a result of their involvement. One study (Kreicbergs et al., 2004) did demonstrate that 28% of participants were negatively affected by their participation, although the paper did not elucidate further as to what these effects were. However 99% still viewed the study as valuable. One paper which explores the incongruity between the perspectives of participants and Research Ethics Boards states this is an important area for further research as the decision of the latter has the potential to influence the experience of the former (Buckle et al., 2010).

To our knowledge, only one other study (Stacey et al., 2009) has explored the experience of parents (both bereaved and controls) participating in stillbirth research, however the study did not assess the anxiety associated with participation. This study aims to explore this gap by assessing the anxiety levels and study acceptability of women participating in a stillbirth case-control study.

Methods

Participants

All participants of the Sydney Stillbirth Study were eligible to participate. The Sydney Stillbirth Study was a population based case-control study investigating risk factors for late pregnancy stillbirth between January 2006 and December 2011. Nine major hospitals in the Sydney metropolitan area participated. Detailed methods have been previously published (Gordon et al., 2015). Briefly, eligible cases were women with a singleton pregnancy who experienced a stillbirth at ≥ 32 weeks gestation. Women were approached by a member of the study team or clinician to participate in the study as soon as possible after receiving the diagnosis of fetal death, taking into consideration the sensitive situation and advice from staff caring for the mother and family. Control women were matched by gestational age and booking hospital, identified through existing hospital databases and randomly selected. They were contacted by a member of the study team either in person or by phone and invited to participate using a standardised pro forma to ensure there was professional courtesy, sensitivity, and consistency in the way the conversation was initiated. Participation in the study included a recorded structured interview intended to be conducted within one week of consenting to the study. The interviews were identical except for two additional questions relating to 'what happened?' prior to the death of the fetus for the case participants. The interview for case participants lasted approximately one to two hours. The interview for the controls lasted approximately 30 minutes. All researchers involved in the interviews observed at least one interview with a bereaved participant conducted by an experienced researcher followed by a period of supervised interviews before they conducted individual interviews.

Data collection

For this follow-up study, identical questionnaires with a cover letter and a stamped, return-addressed envelope were posted to all participants of the Sydney Stillbirth Study in February 2012. This comprised a total of 295 women: 103 cases and 192 controls. Participant study numbers from the original Sydney Stillbirth Study were used in order to link all pre-collected participant's birth and demographic data. All data was entered in a password protected database, and participants were identified only by the study number. For non-respondents, it was planned that at least two attempts would be made to contact participants by phone and/or email.

The questionnaire was designed with multidisciplinary input from clinicians and researchers with expertise in qualitative research methodology, and pilot tested by relevant clinicians, bereaved parents and perinatal epidemiologists. The questionnaire (see Appendix A) included items assessing how women were initially approached: whether it was in person or by phone and who made the initial contact based on the following choices: a doctor, a midwife/nurse, a study researcher, or an option for 'other'. Anxiety and acceptability were assessed using a five point Likert scale ranging from Strongly Disagree to Strongly Agree. Anxiety was evaluated by determining: (1) anxiety at the time of consent, and (2) increased anxiety as a result of participating in the interview. Acceptability of participation was based on: (1) the manner of the person who first approached them about the study, (2) adequate explanation about the research, and (3) overall satisfaction with participating.

For the analysis, responses were grouped into 'Strongly Agree/Agree', 'Neutral', and 'Strongly Disagree/Disagree'. The Spielberger State-Trait Anxiety Inventory (STAI-6) was used to assess the level of anxiety at the time of completing the questionnaire. This short form of the original STAI has been validated for use with bereaved families and is one of the most frequently used measures of anxiety (Marteau and Bekker, 1992; Tluczek et al., 2009). Open ended questions regarding their reasons for participating were asked and there was space for additional comments.

We defined participants as having 'adverse social circumstances', when some of their demographic variables could be broadly defined as factors that had the potential to impact on engagement in health services and completion of the follow up questionnaire. Included factors were: recent immigration to Australia, recently separated or divorced, substance use or maintenance programs.

Statistical analysis

Quantitative data are presented using descriptive statistics. Differences between characteristics of those who responded and those who did not by case or control status were compared using the χ^2 test for categorical variables and the independent *t*-test for continuous variables. The STAI-6 score was compared with the timing of interview following the diagnosis of stillbirth for cases using ANOVA. Text responses to open-ended questions were analysed using thematic analysis (Daly et al., 1997) by two members of the team (DB and AG). The process included: familiarisation with the data (reading and rereading the written responses), independently coding the data using the study objectives and emergent themes and developing a conceptual framework by clustering themes together to best explain the data. Discussion between the researchers continued until there was a consensus of themes. Quotations that directly related to the identified themes or the aims of this study were selected for presentation. Quantitative data were analysed using SPSS version 21.

Ethics approval was given by the Northern Sydney Local Health District Human Research Ethics Committee (Study ID: 0605-081M) and informed consent obtained from all participants.

Findings

A total of 35 (34%) case participants and 65 (33.8%) control participants returned the completed questionnaire (see Fig. 1). One additional case questionnaire was returned incomplete and therefore was only included in the demographic data and not in the analysis. One participant completed the questionnaire but did not complete the STAI-6. Two phone and/or email attempts were made to contact the case participants. One case participant opted to complete the questionnaire by phone. Only one attempt was made to contact a small group of control participants as saturation of thematic analysis was reached well before collecting all the responses.

Characteristics of respondents

Socio-demographic differences between those women who participated in this follow-up study by case or control status compared to those who did not respond are shown in Table 1. There were no significant differences in sociodemographic factors between cases and controls who responded to the questionnaire. Although the average time from recruitment to completion of the questionnaire was longer for case participants with a mean time of 26 months (sd 15.9) compared to controls with a mean time of 20 months (sd 15.6), this difference was not statistically significant ($p=0.069$). Differences were noted between case and control participants who did not respond to the questionnaire. The controls were significantly more likely than cases to be in private care compared to public care (26.8% versus 11.9%, $p=0.017$), living with a partner at the time of birth compared to not living with a partner (94.5% versus 82.1%, $p=0.006$) and tertiary educated compared to completing high school or less (71.7% versus 44.8%, $p < 0.0001$). The cases were significantly more likely than controls to not be in paid work as compared to those working part time or full time or on educational leave (35.8% versus 12.6%, $p < 0.0001$). These differences in maternal characteristics reflect the overall differences in the women who participated in the Sydney Stillbirth Study (Gordon et al., 2015).

Anxiety

Over half of the cases and controls stated they did not feel anxious at time of consent or as a result of participation (see Fig. 2). The majority of participants in both groups reported that participation in the study did not increase their anxiety (57.1% of cases and 89.2% of controls). A similar number of cases (14.3%) and controls (15.3%) agreed that they were anxious at the time of consenting to the study. Although there was an increased proportion of cases who reported that participation in the research increased their anxiety (14.3% to 22.8%) this increase was not statistically significant. However, there was a statistically significant increase in controls who reported that participation did not increase their anxiety (56.9% versus 89.2%, $P = < 0.001$). Over 60% of the case participants were interviewed within a week of their stillbirth. Anxiety response was not affected by timing of interview ($F=1.2$; $P=0.37$). Case participants had significantly higher STAI-6 scores at the time of completing the questionnaire with a mean score of 12.47 (sd 4.05) compared with 8.77 (sd 2.79) for controls ($p < 0.0001$).

Experience of recruitment

All of the case participants were contacted directly in person to participate in the Sydney Stillbirth Study. Almost half (45%) of the control participants were approached directly in person, with the rest (55%) by phone. All but one of the case participants agreed or strongly agreed that the manner of the person approaching them was adequate (see Fig. 3).

Acceptability

Over 85% of all participants agreed/strongly agreed that the study was acceptable regarding manner of approach, explanation of the study, and overall satisfaction with participation (see Fig. 3). The one case participant who 'strongly disagreed' with the manner of approach, also 'strongly agreed' that she was satisfied with participating and stated that she would be willing to be contacted

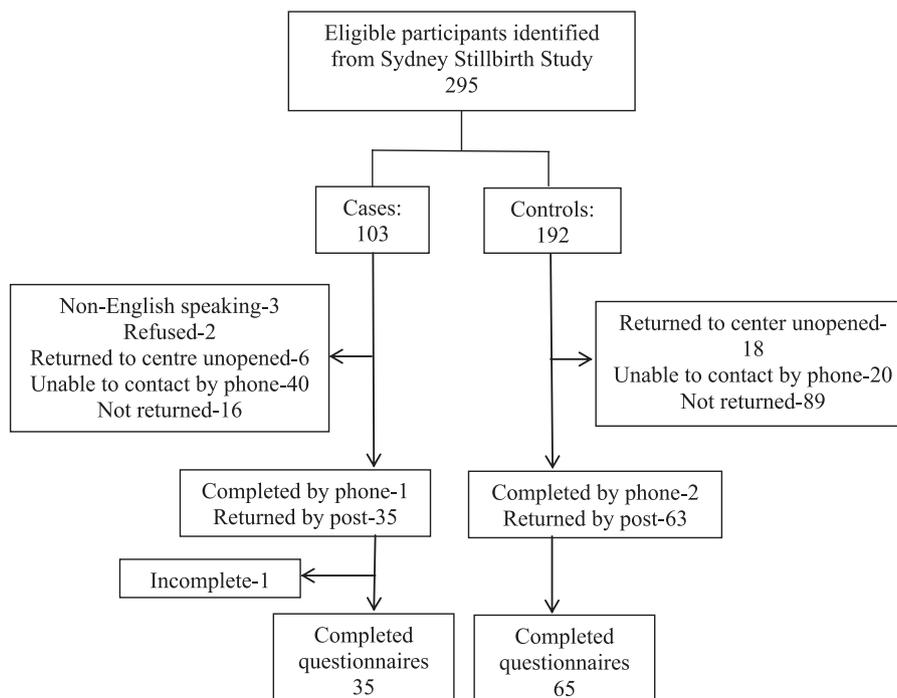


Fig. 1. Study flow chart of eligible participants who returned completed questionnaires.

Table 1
Characteristics of Responders by Case/Control Status.

Maternal characteristics	Responders n = 101 (%)		p	Non-responders n = 194 (%)		p
	Cases 36 n%	Controls 65 n%		Cases 67 n%	Controls 127 n%	
Maternal age						
< 35	23(63.9)	39(60.0)	n.s*	50(74.6)	82(64.6)	n.s.
35–39	10(27.6)	17(26.2)		12(17.9)	36(28.3)	
≥ 40	3(5.3)	9(13.8)		5(7.5)	9(7.1)	
Primiparous	16(44.4)	37(56.9)	n.s.	37(55.2)	67(52.8)	n.s.
Private care	14(38.9)	25(38.5)	n.s.	8(11.9)	34(26.8)	0.017
Not in paid work	2(5.6)	2(3.1)	n.s.	24(35.8)	16(12.6)	< 0.0001
Living with partner	36(100.0)	63(96.9)	n.s.	55(82.1)	120(94.5)	0.006
Smoking	3(8.3)	7(10.8)	n.s.	11(16.4)	18(14.2)	n.s.
Adverse social	1(2.8)	4(6.2)	n.s.	11(16.4)	10(7.9)	0.069
Tertiary education	30(83.3)	52(80.0)	n.s.	30(44.8)	91(71.7)	< 0.0001
Born overseas	14(38.9)	25(38.5)	n.s.	30(44.8)	70(55.1)	n.s.
Drugs in pregnancy	0(0.0)	3(4.6)	n.s.	3(4.5)	1(0.8)	n.s.

* n.s: non-significant.

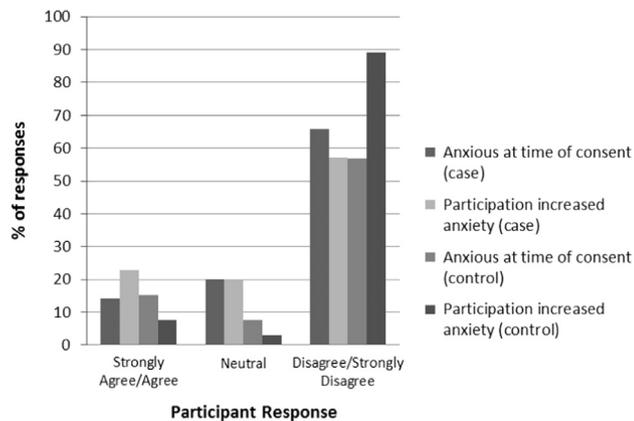


Fig. 2. Anxiety at time of consent and after participation in stillbirth study.

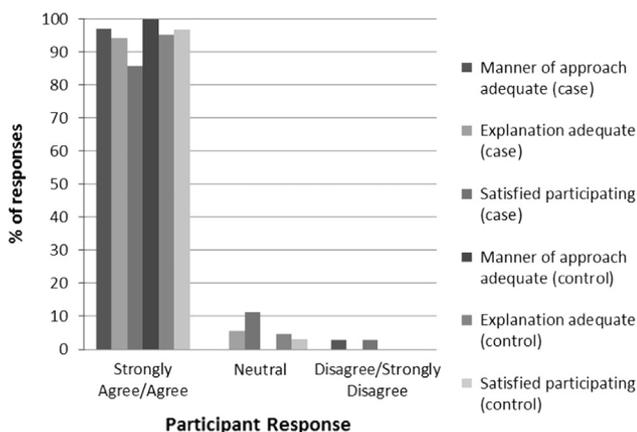


Fig. 3. Acceptability related to manner of approach, explanation of research and participation in stillbirth study.

for further research. The only case participant who was dissatisfied with her participation in the study also indicated considerable dissatisfaction with her general clinical care during her time in hospital.

Qualitative comments

Written responses regarding reasons for participation were initially categorised into themes specific to cases and controls. As

this analysis resulted in identical themes other than an extra theme for control participants, the final results were categorised into three main themes: altruism, finding answers, and attributing meaning with an additional sub-theme identified for control participants only: ease of participation.

'Altruism': The majority of case participants and all the control participants stated they participated in order to prevent this tragedy from recurring to themselves or to others. Several stated they wished they could have contributed more:

'Being part of this study helped with my grief - helped feel I could be involved in a small way in helping prevent stillbirths in the future.' (S022)

'I am buoyed by the idea that this work may contribute to another woman not having to go through the horror of late-term stillbirth.' (S088)

'I agreed to the study hoping I could help prevent stillbirth. I hope any information gathered from me and my pregnancy could help at least 1 mother.' (C155)

Written comments suggest that even when anxiety was present, it did not appear to hinder their willingness to participate:

'I wrote 'sometimes' for 'tense' and 'worried', but am happy to send you any other information that may be of help. Thank you.' (S078)

'Even though I've said I was anxious when asked to participate and during the interview, I was still more than happy to go through with it. The anxiety was simply brought to the fore because of discussing it. I believe it would be present in any expectant mother's mind.' (C059)

'Finding Answers': Searching for answers as to why the death occurred was a common theme, especially for the bereaved participants. Many controls had friends who had experienced this type of loss or worked in areas related to clinical care, research or support groups, making the study relevant to their personal experience and fostering their desire to help find solutions:

'Medical background - belief in need for quality research. Wanting to help prevent stillbirth if possible.' (C186)

'Desperate need to find out why my baby died. It was empowering. I wanted to participate more than I was asked to.' (S030)

'Way of giving back. Friend had stillbirth - wanted to help.' (C025)

'Attributing Meaning': Many comments were made about the importance of participating in order to attribute meaning to the stillbirth. For bereaved mothers this particularly meant giving

purpose to their baby's life. Many felt their participation helped with their grief. Both groups of participants commented on hoping to raise awareness about stillbirth and improving care:

'It helped us feel that (our baby's) life meant something, that he made a contribution, no matter how small.' (S100)

'I feel it's an important issue and one that I know very little about even after having 3 kids.' (C180)

'So that there is a chance something positive might come out of the death of our daughter - even if it is just how you might be treated at the time.' (S071)

'Ease of Participation': Many control participants stated the study was easy to participate in secondary to the approachable manner of the researcher and the undemanding nature of the study:

'Made me feel so comfortable. No obligation. Interviewer manner so friendly and personable.' (C025)

'It was easy without me having to make appointments for tests. I felt it was a very important study and more research is needed.' (C191)

'Good to help out. Good reassurance that I could pull out at any time. No testing on baby. No imposition.' (C168)

Discussion

This is the first time a study has examined both anxiety and acceptability levels of pregnant and bereaved women participating in a stillbirth study. The bereaved participants consented very soon after being informed that their pregnancy had resulted in a fetal death, and the controls participated while still pregnant with a live baby in their last trimester of pregnancy. Our results show that there was a high level of acceptability and satisfaction among both groups of individuals as a result of their participation in the Sydney Stillbirth Study. There was a non-significant increase in anxiety reported in one fifth of the case participants and a significant increase in the number of control participants who reported that participation did not increase their anxiety. The timing of interview in respect to the fetal loss did not affect their anxiety.

Despite concerns from hospital staff and ethics committees about the timing of recruitment approach, 87% of the cases and 86% of the controls consented to take part in the Sydney Stillbirth Study (Gordon et al., 2015), demonstrating a high level of participation acceptability. A previous study highlights the discrepancies among researchers and even the bereaved themselves as to what constitutes the most appropriate time to approach for consent (Beck and Konnert, 2007). Discerning between the emotional response to grief and the competency to understand the nature of the study with its potential risks and benefits was an important factor. A large majority of the participants in that study (85.7%) reported that they were able to make an informed decision to consent soon after the death (Beck and Konnert, 2007). This correlates with the number of participants who consented to the Sydney Stillbirth Study.

Acceptability can also be affected by the manner of recruitment. While one study indicated that bereaved parents valued an informative letter about the study as being less intrusive (Hynson et al., 2006), the nature of the Sydney Stillbirth Study and its early recruitment to minimise recall bias precluded this method. Furthermore, the research team felt direct contact with the participants was a more personal method. Qualitative comments as to the professionalism, warmth, and empathy exhibited at the initial approach, as well as the low levels of anxiety documented in this follow-up study relating to the time of consent suggest that this method was acceptable.

Likewise, over 85% of participants indicated a high level of satisfaction with their participation. This is likely due to the altruistic motives for participating as well as the continued assurance that participants could withdraw at any point during the course of the study or questioning (Burnell and O'Keefe, 2004). Although the bereaved participants found the questions relating to 'What happened before the death of your baby?' difficult and were often tearful while recounting their story, at no point did anyone choose not to continue, despite assurance that this was an option. On the contrary, the qualitative results suggest that many of the bereaved parents felt 'empowered' and grateful to describe their experience. Others have also suggested that there are positive benefits for bereaved parents when they are able to recount their experience with an interested and engaged researcher (Buckle et al., 2010; Brabin and Berah, 1995). This was also anecdotally observed by our research team, who reported that engagement by the hospital staff improved over time and they became more receptive to contacting the team about eligible participants as they witnessed the positive responses by the participants.

The significant increase in the number of controls who did not experience a higher level of anxiety as a result of their involvement in the study invalidates concerns about participation of healthy pregnant women in stillbirth research. Their comments suggest that the beneficence of participation combined with the warmth and professionalism of the research team positively outweighed any anxiety that may have been present.

Strengths and limitations

Strengths of this study include using a validated anxiety scale and pilot testing of the questionnaire by bereaved parents to assess acceptability. Furthermore, this study is unique as it assesses the impact of participation on bereaved individuals who were approached close to the time of their loss. Likewise, it assesses the responses of pregnant women being approached about a sensitive topic, showing the feasibility of conducting stillbirth research on healthy people at what is perceived to be a particularly vulnerable time. However, the small sample size owing to the low response rate makes it difficult to generalise our results to all participants of stillbirth research. There were socio-demographic differences between cases and controls only in those who didn't respond to the questionnaire, which is consistent with previous literature indicating that the socially disadvantaged are less likely to participate (Burnell and O'Keefe, 2004). It should also be noted that there were similar response rates for both cases and controls potentially indicating that the reasons for non-response are those common to surveys rather than secondary to emotional stress which would be predictably higher in bereaved participants. Although it is possible that the questionnaire may have triggered an emotional response in bereaved mothers making her less likely to respond, there was also no difference in the qualitative analysis between the two groups, despite the cases showing a higher STAI-6 level at the time of completing the questionnaire. Hospital records confirm that during the six years it took to complete the study, many participants had moved house or changed contact details and were unable to be contacted. This is consistent with other studies in pregnant populations (Raynes-Greenow et al., 2008). Unfortunately, at the start of the study, we did not have the resources to consent for the use of mandated state health information as a means of contacting participants at a later date. Studies performed in countries that allow for research contact via mandated registries (e.g. Sweden) show a higher response rate (Rådestad et al., 2009). Despite these limitations, our response rate is similar to other studies using postal questionnaires to assess stillbirth experiences (Stacey et al., 2009; Gravesteen et al., 2013).

The time between participation and the follow-up questionnaire may also have affected recall about feelings of participation. However, the direction of this effect is uncertain, as had participation caused anxiety, the questionnaire may have triggered an instant recall of the stress experienced at the time. Conversely, a longer length of time may have given a more objective perspective, allowing the participants to process their grief with improved coping mechanisms (Scott et al., 2002).

Conclusions

This is one of the first studies to assess both bereaved and pregnant women's responses to stillbirth research. We have shown that participation in the Sydney Stillbirth Study resulted in high levels of participant acceptability and satisfaction amongst both cases and controls and, while there was a slight increase in anxiety among case participants as a result of their participation in the study, the increase was not statistically significant, affected only one in five cases and did not appear to affect satisfaction. The majority of both groups reported that study participation did not increase their anxiety with reasons for participation centred around finding answers, altruism and attributing meaning. Our results are consistent with previous studies showing a positive response to participation in bereavement research. An empathic and professional approach was appreciated by the participants. Findings from this study are reassuring for 'gatekeepers' who make decisions about women's participation in fetal loss research, and suggests that 'protecting' families may require further justification.

Role of the funding source

The primary author for this manuscript declares that funding for her role as researcher in this project was received by the Stillbirth Foundation Australia, which did not contribute in any way to the design, data collection, analysis, writing or publication of this manuscript.

Conflict of interest

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

Acknowledgements

The authors are grateful to the Stillbirth Foundation Australia for their generous funding of the Sydney Stillbirth Study which contributed to this follow up study. We would like to thank A/Professor Natasha Nassar and Ms. Shelley Reid for their invaluable assistance in the development of the questionnaire. Most importantly, we acknowledge the amazing families who contributed to this study – the honesty and sincerity reflected in their responses are greatly appreciated.

Funding for this project was received from the Stillbirth Foundation Australia, who did not contribute in any way to the design, data collection, analysis, or writing of this manuscript.

Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at <http://dx.doi.org/10.1016/j.midw.2015.07.005>.

References

- Beck, A.M., Konnert, C.A., 2007. Ethical issues in the study of bereavement: the opinions of bereaved adults. *Death Stud.* 31, 783–799.
- Brabin, P.J., Berah, E.F., 1995. Dredging up past traumas: harmful or helpful? *Psychiatry Psychol. Law* 2, 165–171.
- Buckle, J.L., Dwyer, S.C., Jackson, M., 2010. Qualitative bereavement research: incongruity between the perspectives of participants and research ethics boards. *Int. J. Soc. Res. Methodol.* 13, 111–125.
- Burnell, R.H., O'Keefe, M., 2004. Asking parents unaskable questions. *Lancet* 364, 737–738.
- Cacciatore, J., 2013. Psychological effects of stillbirth. *Semin. Fetal Neonatal Med.* 18, 76–82.
- Cook, A.S., 1995. Ethical issues in bereavement research: an overview. *Death Stud.* 19, 103–122.
- Daly, J., Kellehear, A., Gliksman, M., 1997. *The Public Health Researcher: A Methodological Approach*. Oxford Uni Press, Melbourne, Australia.
- Dent, A., Condon, L., Blair, P., Fleming, P., 1996. A study of bereavement care after a sudden and unexpected death. *Arch. Dis. Child.* 74, 522–526.
- Dyregrov, K., 2004. Bereaved parents' experience of research participation. *Soc. Sci. Med.* 58, 391–400.
- Gordon, A., Raynes-Greenow, C., Bond, D., Morris, J., Rawlinson, W., Jeffery, H., 2015. Sleep position, fetal growth restriction, and late-pregnancy stillbirth: The Sydney Stillbirth Study. *Obstet. Gynecol.* 125, 347–355.
- Gravesteen, I., Helgadóttir, L., Jacobsen, E.-M., Rådestad, I., Sandset, P.M., Ekeberg, Ø., 2013. Women's experiences in relation to stillbirth and risk factors for long-term post-traumatic stress symptoms: a retrospective study. *BMJ Open* 3, 1–10.
- Hynson, J., Aroni, R., Bauld, C., Sawyer, S., 2006. Research with bereaved parents: a question of how not why. *Palliat. Med.* 20, 805–811.
- Kelly, M., Trinidad, S., 2012. Silent loss and the clinical encounter: parents' and physicians' experiences of stillbirth – a qualitative analysis. *BMC Pregnancy Childbirth* 12, 137, 1–15.
- Kreicbergs, U., Valdimarsdóttir, U., Steineck, G., Henter, J.-I., 2004. A population-based nationwide study of parents' perceptions of a questionnaire on their child's death due to cancer. *Lancet* 364, 787–789.
- LaRoche, C., et al., 1984. Grief reactions to perinatal death: a follow-up study. *Can. J. Psychiatry* 29, 14–19.
- Marteau, T.M., Bekker, H., 1992. The development of a six-item short-form of the state scale of the Spielberger State–Trait Anxiety Inventory (STAI). *Br. J. Soc. Clin. Psychol.* 31, 301–306.
- Rådestad, I., Steineck, G., Nordin, C., Sjogren, B., 1996. Psychological complications after stillbirth—influence of memories and immediate management: population based study. *BMJ* 312, 1505–1508.
- Rådestad, I., Surkan, P.J., Steineck, G., Cnattingius, S., Onelöv, E., Dickman, P.W., 2009. Long-term outcomes for mothers who have or have not held their stillborn baby. *Midwifery* 25, 422–429.
- Raynes-Greenow, C.H., Nassar, N., Roberts, C.L., 2008. Residential mobility in a cohort of primiparous women during pregnancy and post-partum. *Aust. N. Z. J. Public Health* 32, 131–134.
- Scott, D.A., Boyle, F.M., Bain, C.J., Valery, P.C., 2002. Does research into sensitive areas do harm? Experiences of research participation after a child's diagnosis with Ewing's sarcoma. *Med. J. Aust.* 177, 507–510.
- Seamark, D.A., Gilbert, J., Lawrence, C.J., Williams, S., 2000. Are postbereavement research interviews distressing to carers? Lessons learned from palliative care research. *Palliat. Med.* 14, 55–56.
- Sque, M., 2000. Researching the bereaved: an investigator's experience. *Nurs. Ethics* 7, 23–34.
- Stacey, T.M.E., Thompson, J., Ekeroma, A., Zuccollo, J., McCowan, L., 2009. The Auckland Stillbirth Study: the experience of participants. *J. Paediatr. Child Health* 45, A182, A10–A71.
- Tluczek, A., Henriques, J.B., Brown, R.L., 2009. Support for the reliability and validity of a six-item state anxiety scale derived from the State-Trait Anxiety Inventory. *J. Nurs. Meas.* 17, 19–28.
- Wild, V., 2012. How are pregnant women vulnerable research participants? *Int. J. Fem Approaches Bioet.* 5, 82–104.