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Anxiety and acceptability related to participation in stillbirth research

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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.

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; Cacciatore, 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 $\geq$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.

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