Does research into sensitive areas do harm? Experiences of research participation after a child's diagnosis with Ewing's sarcoma

Debbie A Scott, Patricia C Valery, Frances M Boyle and Christopher J Bain

ABSTRACT

Objective: To investigate family members' experiences of involvement in a previous study (conducted August 1995 to June 1997) following their child's diagnosis with Ewing's sarcoma.

Design: Retrospective survey, conducted between 1 November and 30 November 1997, using a postal questionnaire.

Participants: Eighty-one of 97 families who had previously completed an in-depth interview as part of a national case–control study of Ewing's sarcoma.

Main outcome measures: Participants' views on how participation in the previous study had affected them and what motivated them to participate.

Results: Most study participants indicated that taking part in the previous study had been a positive experience. Most (n = 79 [97.5%]) believed their involvement would benefit others and were glad to have participated, despite expecting and finding some parts of the interview to be painful. Parents whose child was still alive at the time of the interview recalled participation as more painful than those whose child had died before the interview. Parents who had completed the interview less than a year before our study recalled it as being more painful than those who had completed it more than a year before.

Conclusions: That people suffering bereavement are generally eager to participate in research and may indeed find it a positive experience is useful information for members of ethics review boards and other "gatekeepers", who frequently need to determine whether studies into sensitive areas should be approved. Such information may also help members of the community to make an informed decision regarding participation in such research.

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SURVEYS IN WHICH people are asked to give information about their health, preferences and behaviours are fundamental to some areas of health research, such as public health. Such research raises ethical considerations that are not always as clear-cut as those of biomedical research or clinical trials, where adverse effects may be more obvious and easier to quantify. In survey research, the possible risks and benefits are essentially of a psychological nature and therefore less tangible or observable. Interview situations can be threatening, particularly when there is uncertainty about what the questions will involve, anxiety about reawakening painful memories or disclosing sensitive information, or concern about the credibility of the interviewer.[1,2] However, a number of researchers have suggested that some people participate in survey research for reasons of altruism[1,2] and that participation can be of personal benefit to the participants.[3,4]

Researchers, ethics committees and research "gatekeepers" frequently judge the likely impact on participants of a particular study on the basis of intuition, past experience (professional and personal) and cultural expectations. Such decisions are seldom easy and there may be diversity of opinion — if this were not the case, there would be no need for ethical review.[5]

Our article describes the experience of participating in a research interview from the perspective of families whose child has had Ewing's sarcoma. Unlike many other studies, which have asked people's views on participation at the time of the survey interview, our study provides a longer-term perspective, allowing people to reflect on experiences some time after their actual study participation.

METHODS

The previous study

The previous study, conducted by members of our research team, was a case–control design (Box 1), details of which are described elsewhere.[6] Of 155 cases identified, data were collected by structured interviews from 132 (85%) between August 1995 and June 1997.

The current study

Ninety-seven of the 132 case interviews in the previous study involved the participation of family members other than the diagnosed child. In November 1997, 3–36 months (average, 14 months) after completion of the previous study interviews, we invited these 97 former participants to complete a mailed, self-administered follow-up questionnaire. The questionnaire contained open- and close-ended questions about how participation in the previous study had affected them and what had motivated them to participate (eg,

For editorial comment, see page 471

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1: A population-based case–control study of Ewing's sarcoma in Australia[6]

- Cases were 132 out of 155 patients (response rate, 85%) under 40 years of age diagnosed with Ewing's sarcoma between January 1991 and June 1996.
- Control subjects were 428 out of 473 people (response rate, 90%) selected randomly from the Australian population by telephone, matched to cases by age group and State of residence at diagnosis.
- Information was obtained by interview for pregnancy-related factors, subjects' medical history, sociodemographic status, family history of cancer, place of residence, as well as detailed information about farms and occupational chemical exposures.
- Of 132 cases, 93 were alive at the time of the interview. Their average age was 14.1 years (range, 0–35 years).

Results showed that parents of children with Ewing's sarcoma were more likely to have worked on farms, but this association was not statistically significant.

"Were the questions painful?"; "Did the questions make you uncomfortable?"; "What were the unexpected questions about?").

We conducted χ^2 analyses using SPSS[7] to determine patterns of association between variables.

Ethical clearance was obtained from the Behavioural and Social Science Ethical Review Committee of the University of Queensland.

RESULTS

Some demographic characteristics of participants and non-participants in our study are shown in Box 2. Of 97 questionnaires sent out, 81 were returned (83.5%), eight families did not return the questionnaire despite a telephone prompt, and eight could not be contacted. Respondents and non-respondents did not differ significantly in terms of socioeconomic status, maternal education, patients' sex or patients' age at diagnosis. However, families of children who had died were significantly more likely to participate in our study than those whose child was living ($P < 0.001$). Almost all participants ($n = 79$ [97.5%]) had entered the previous study with the belief their participation would be beneficial to others (Box 3). Most were pleased to be involved in the current study ($n = 76$ [93.8%]), despite almost half anticipating the interview would be painful ($n = 38$ [46.9%]).

Parents of a child who had died before the interview were significantly less likely to expect that answering questions about their child's life before diagnosis would be painful than those whose child was alive at interview (1 of 47 [two responses missing] compared with 17 of 32; $P = 0.02$). Some responses to open-ended questions provided insight into this finding: "The interview was painful, but if it saves one child from suffering as my daughter had to her death will not have been in vain."; "We particularly liked the interview, for the chance to sit and talk face to face with a wonderful lady who was professional, compassionate, understanding and answered all our questions honestly."

Most participants had felt in the previous study that they could refuse to answer questions that made them

2: Demographic characteristics of participants and non-participants in our study, assessed in relation to the previous study

	Number (%) of participants ($n = 81$)	Number (%) of non-participants ($n = 16$)
Status of patient (child with Ewing's sarcoma) at time of interview		
Dead	49 (60.5%)	2 (12.5%)
Alive	32 (39.5%)	14 (87.5%)
Interviewee(s)		
Mother only	37 (45.7%)	6 (37.5%)
Father only	4 (4.9%)	2 (12.5%)
Mother and father	26 (32.1%)	3 (18.8%)
Mother and patient (± father)	9 (11.1%)	4 (25.0%)
Parent(s) and siblings (± patient)	4 (4.9%)	0
Mother and friend	1 (1.2%)	1 (6.3%)
*Time elapsed between interview in previous study and current study**		
12 months or less	37 (45.7%)	8 (50.0%)
More than 12 months	44 (54.3%)	8 (50.0%)
Time from child's diagnosis with Ewing's sarcoma to participation in previous study†		
≤6 months	10 (12.3%)	1 (6.3%)
7–20 months	14 (17.3%)	4 (25.0%)
21–48 months	35 (43.2%)	10 (62.5%)
49 or more months	18 (22.2%)	1 (6.3%)
Age of child at diagnosis		
0–4 years	9 (11.1%)	1 (6.3%)
5–9 years	16 (19.8%)	2 (12.5%)
10–14 years	30 (37.0%)	9 (56.3%)
15–19 years	20 (24.7%)	4 (25.0%)
20+ years	6 (7.4%)	0
Mother's education‡		
Primary school only	2 (2.5%)	0
Incomplete high school	38 (46.9%)	10 (62.5%)
Completed high school or apprenticeship	20 (24.7%)	4 (25.0%)
University or college of technical and further education	19 (23.5%)	2 (12.5%)

*Range, three months to three years. †Four responses missing. ‡Two responses missing.

3: Feelings about participation in the previous study reported by the 81 participants in our current study

Questionnaire item	Number (%) answering "yes"	Number (%) answering "neutral"	Number (%) answering "no"
Felt eager to participate	76 (93.8%)	4 (4.9%)	1 (1.2%)
Were glad they participated	79 (97.5%)	2 (2.5%)	0
Felt the questions were as expected	45 (55.6%)	32 (39.5%)	4 (4.9%)
Felt that a benefit of participation was that they could talk about their child's illness	52 (64.2%)	25 (30.9%)	4 (4.9%)
Felt they could refuse to answer questions that made them uncomfortable	72 (88.9%)	8 (9.9%)	1 (1.2%)
Would have participated in a similar study as a control*	57 (70.4%)	21 (25.9%)	3 (3.7%)
Felt obliged to participate	7 (8.6%)	10 (12.3%)	64 (79.1%)
Expected interview to be painful	38 (46.9%)	33 (40.7%)	10 (12.3%)
Questions made respondent(s) wonder if they could have prevented the child's illness	18 (22.2%)	15 (18.5%)	48 (59.3%)
Felt that interview was more painful than they anticipated	6 (7.4%)	17 (21.0%)	58 (71.6%)
Felt that their participation would be beneficial to others	79 (97.5%)	2 (2.5%)	0
Would recommend to others in their position that they participate in similar studies	74 (91.4%)	5 (6.2%)	2 (2.5%)

* That is, a healthy participant in a case–control study.

uncomfortable. Nine (11.1%) agreed some questions had made them feel uncomfortable; seven (8.6%), including five whose child was alive at the previous study interview, had found the interview *in general*, rather than any particular question, was disquieting.

Most participants believed their participation in the previous study benefited others; two-thirds felt participation was personally beneficial because they could talk about their child's illness. Some ($n = 6$ [7.4%]) had found the interview more painful than expected. Participants who had been interviewed one year or less before our study ($n = 15$ [40.5%]) reported finding the interview more painful than those who had been interviewed more than a year before our study ($n = 7$ [16.7%]) ($P = 0.02$). No participant disagreed with the statement, "I am glad to have participated in the interview".

Almost half the participants ($n = 35$ [43.2%]) believed the interview had produced some good from an otherwise bad situation, although a substantial number ($n = 22$ [27.2%]) felt the study had not affected them. Others ($n = 9$ [11.1%]) said the interview had encouraged discussion or that they had found it thought-provoking ($n = 3$ [3.7%]). No family felt that participation in the study had "upset them".

DISCUSSION

The very nature of this study raises concerns about possible sample bias. Although the response rate was high, at least some non-respondents may have elected not to participate in our study because their experience of participation in the previous study was less than positive. Families of children who were alive at the time of the previous study were significantly less likely to participate in our current study and found participation in the previous study more painful — if the views of non-respondents had been included, there may have been more variation in the responses concerning perceptions of the interview.

The results of a small number of studies addressing related issues of grief and loss are generally consistent with our findings.[3,4,8-12] Neugebauer et al,[3] investigating depressive symptoms in women who had miscarried, discovered that women who had completed interviews at two weeks, six weeks and six months after miscarriage had lower depression levels than those who had completed an interview only at six weeks and six months. This was attributed to "unintentional therapeutic and test effects of study interviews".[3] Kitson et al,[8] investigating the effects of violent deaths on families, noted that the majority of participants in their study felt it was useful to talk to someone, even though participation was distressing. In research on how terminally ill people changed their lives once they discovered they were dying, Kellehear[4] wrestled with the fact his questions seemed to leave participants in tears. He was concerned that this may be harmful; however, many said being able to review their lives was beneficial. Similar sentiments were found in a study examining next-of-kin attitudes of people participating in a case–control study of adult leukaemia in Seattle, USA.[9]

Our results support these findings, despite some participants expecting and finding the original interview to be painful. Almost all were glad to have participated, as they felt that the research gave them the opportunity to discuss their child's illness and that their participation would benefit others.

The findings of our study provide useful information for members of ethics review boards who consider whether a study should be approved, and for other "gatekeepers" who decide whether researchers should be granted access to specific populations (ie, patients and families). It is also useful for researchers designing and conducting studies into sensitive and painful areas, who must consider the needs and expectations of those who participate in the research. It is important to note that interviewers need to be skilled and properly trained, as results may be affected by the skills of interviewers. Finally, our study provides potentially useful information for members of the community who may be invited at some stage to participate in research.

The risks and benefits of participation in survey research, particularly when it deals with sensitive or painful topics, are real but often intangible. Many people accept these risks and participate in research they feel is worthwhile and

beneficial to others[1] and these results have been echoed in other studies.[3,4,8-12] Overall, people are pleased to participate, despite anticipating possible distress. Professionalism, interpersonal skills, compassion, and awareness of the potential for negative experiences from participants are essential components in ensuring that participation in research "does no harm". Provided these and other established ethical guidelines are met, there is a strong case for giving people the opportunity to decide whether the benefits outweigh the risks in their own particular situation.

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COMPETING INTERESTS

None identified.

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OBITUARIES

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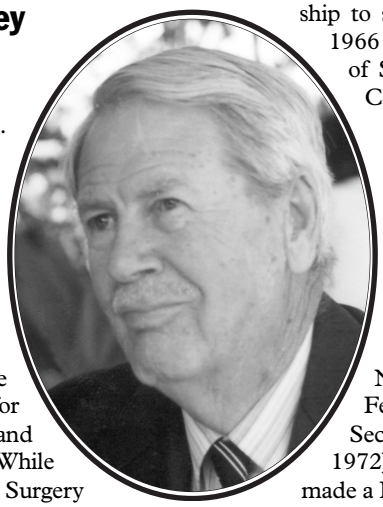

TED STUCKEY epitomised "quiet achievement". Born on 15 June 1908, he grew up in Inverell, in northern New South Wales, where he was dux of his school. Later, as a medical student living at St Andrew's College, Sydney University, he excelled academically and in sport. He represented the College in rowing, and played hockey for the College, the University, and a combined Australian universities' team.

After doing his residency at the Royal Prince Alfred Hospital and the Royal Alexandra Hospital for Children (RAHC), Ted married Joan Vowell and moved into general practice in Scone, NSW. While working in this practice he obtained his Master of Surgery degree.

Ted returned to Sydney in 1939 to become a paediatric surgeon, and was appointed Honorary Relieving Assistant Surgeon at RAHC. When war intervened, he joined the Field Ambulance Service. He served until late 1944 in Queensland, then New Guinea, becoming second-in-charge of the 111th Casualty Clearing Station and attaining the rank of Major.

From 1945, as Honorary Assistant Surgeon at RAHC, Ted and his colleagues did pioneering work in cardiothoracic and abdominal surgery. Ted's brother Doug was also part of the Congenital Heart Disease team that was involved in the early development of cardiac catheterisation and angiocardiography.

In 1948, Ted gained his Fellowship of the Royal Australasian College of Surgeons. In 1958 he was awarded a Fulbright scholarship to study at Harvard Medical School. From 1958 to 1966 he lectured in paediatric surgery at the University of Sydney. He continued at RAHC as an Honorary Consultant Surgeon until 1973.

In later years, Ted adopted a more relaxed lifestyle, doing sessional work with the Commonwealth Health Department until 1988 and Surgical Assistant work until 1994 (then aged 86!).

Ted was a founding member of the Medical Benefits Fund in 1945 and served on its Council until 1971. He was also heavily involved with the Australian Medical Association. He was a member (1953–1966) and president (1961–1962) of the NSW Branch Council; a member of the AMA Federal Council (1964–1966); Assistant General Secretary, then Deputy Secretary General (1966–1972); and Secretary General (1972–1973). He was made a Fellow of the AMA in 1964. He was secretary of the AMA/benefit fund working party, which produced a plan for a voluntary health insurance scheme that was largely adopted by the federal government and introduced in 1970. He was also a member of the Medical Benefits Schedule Advisory Committee.

Although deeply committed to his profession, Ted remained a devoted husband and father to his five children. Over the years, he built for his family a swimming pool, a terraced garden with a badminton court, and three unique folding caravans in which he loved to take them on camping holidays.

Ted died on 7 June 2002 of acute renal failure.

Michael E V Stuckey