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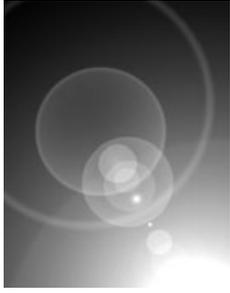
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ORIGINAL ARTICLE

Doing research on people with learning disabilities, cancer and dying: ethics, possibilities and pitfalls

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Accessible summary

- We have done research about cancer, death and dying. People with learning disabilities who had cancer were in our studies. This paper is about making sure that our research is *ethical*. This means that we don't want to cause any harm (or make people upset) when we do the research. We ask:
- How do we find people to be in our studies?
- What should we do if people cannot understand the research they are in?
- What should we do if people do not want us to use a made-up name in our writing?
- What should we do if, during the research, we see that people with learning disabilities are not treated right?
- What is the relationship with the researcher? Is the researcher a nurse, a helper or a friend?

Summary

There has been increasing recognition of the fact that people with learning disabilities can and should be involved as active participants in research. However, in the area of cancer and palliative care, they continue to be protected and excluded from contributing to expanding our knowledge base and the shaping of care provision. This paper explores the issues involved in including people with learning disabilities as participants in research around cancer, death and dying. It stems from the authors' own experience of conducting research around cancer and palliative care, where people with learning disabilities were the key informants. Methodological issues include the choice of methodology and problems around recruiting a sample. Ethical issues include informed consent, participants' requests to forego anonymity, observation of sub-optimal care and issues around the boundaries between the roles of researcher and clinician.

Keywords Cancer, death and dying, ethics, learning disabilities, qualitative research, research methodology

Introduction

This paper considers the methodological and ethical issues encountered in including people with learning disabilities in research around cancer, death and dying. The term 'inclusive research' has been used in different ways in the literature, ranging from research where people with learning disabilities are involved as research participants, to research where they are part of the planning or conducting of the research, either as advisors or as active researchers. Although people with learning disabilities were indeed members of the research team in our studies (from concept through to data analysis), this paper focuses on the pitfalls and problems we encountered when *the population to be studied* also consisted of people with learning disabilities who had cancer, and, in some cases, were dying. We hope that sharing our experiences will help other researchers in preparing to conduct research around this or similar important but highly sensitive topics.

Including people with learning disabilities in research is increasingly recognised as important, by people with learning disabilities themselves as well as academics (Department of Health 2006; Gilbert 2004; Owen 2001; Walmsley 2001, 2004; Williams 1999). The premise that it was important and feasible to include people with learning disabilities as active research participants, rather than passive subjects, was a revolutionary idea in the 1980s (Flynn 1986) but has since become the accepted norm for service research. However, inclusion of people with learning disabilities as participants in research around the most sensitive topics remains controversial (Lindop 2006). There are difficult questions about information, consent and the possibility of inflicting harm.

Inclusive research about cancer, death and dying remains a largely uncharted territory. Apart from our own work, we found only two published studies on those topics (up to 2006) that include interviews with people with learning disabilities. Brown *et al.* (2002) do not give explicit details of their interviews with three people with learning disabilities. Todd (2004) describes interviews with 12 people with learning disabilities as part of a larger study of death and learning disability.

There has been ongoing debate within palliative care research generally about whether it is ethical for a vulnerable population of people who are dying to be included in research at all, given that there may be no benefit to compensate the loss of their already limited time and energy (Addington-Hall 2002; Wilkie 2001). Increasingly, it is recognised that participation in research can be an empowering experience for people who are terminally ill, and provide an opportunity to make a final valuable contribution (Hopkinson & Wright 2005; Terry *et al.* 2006). The question is less stark for patients who have cancer and are not terminally ill, but ethical issues remain around the

demands research participation could place on very sick people.

However, a sound body of research is needed to provide an evidence base for sensitive and appropriate support for people with cancer and/or terminal illness. We need to know how the illness is experienced, what helps, what hinders, how we can best support people. The question, therefore, is not *if* we should include people with learning disabilities in research, but *how*. An important and recurrent finding from our studies has been that it is possible for people with learning disabilities to contribute to an expansion of knowledge through participation in research, even when that involves very frightening topics like cancer, death and dying (Tuffrey-Wijne *et al.* 2006, 2007). This echoes Todd's (2004) findings who reported that the people with learning disabilities he interviewed on the topic of death 'turned out to be more relaxed and eager to talk about this issue than I had anticipated' (p. 13). Our own experience of research participants with learning disabilities suggests that they were enthusiastic, not only because of the empowering experience of being listened to, but also because of an understanding that through participation they could make a contribution that would help their peers.

The main research we have drawn on for this paper is 'The Veronica Project', an ethnographic study into the experiences of people with learning disabilities who have cancer (Cresswell & Tuffrey-Wijne 2008; Tuffrey-Wijne & Davies 2007). The aim in ethnography is to understand the world of the group of interest, typically using qualitative participant observation. One of us (IT) was the participant observer. She supplemented and validated her field observations by a triangulation with other data, including unstructured interviews with participants, family members, support workers and healthcare professionals; attendance of case reviews; and studying records. This methodology meant that we were able to include participants who lacked both verbal ability and understanding of their illness; six participants had severe or profound learning disabilities and severe communication difficulties. Two had no speech; four had limited speech that was, at times, hard to understand. In total, 13 people with a cancer diagnosis, and with learning disabilities ranging from mild to severe, were visited regularly by IT (the principal researcher) during time frames ranging from a week to over a year. At the time of writing, seven of these 13 participants have died, three are living with an on-going terminal illness, and three are in remission, hopefully cured. The study will be completed in December 2008. Before and during the 'Veronica Project', we conducted other, smaller studies around cancer, palliative care and learning disabilities, some of which included people with learning disabilities as participants. These have also contributed to our understanding of the pitfalls and possibilities of this kind of research.

We obtained approval for each study from the relevant research ethics committee (COREC for the Veronica Project); we also ensured that the study design was rigorously peer-reviewed. Our studies were guided by research advisory groups which included professional experts and people with learning disabilities.

Methodological issues

Choosing appropriate methodologies

The research topics of cancer, death and dying are highly complex. [McCarthy \(1998\)](#) argues that choosing the appropriate research methodology for the task is itself an ethical issue. [Lindop \(2006\)](#) agrees that 'research methods associated with sensitive research among people with learning disabilities need to be carefully selected so as to be ethically appropriate and unobtrusive' (p. 163).

In our studies, several factors were considered in selecting the most appropriate methodology. How much is already known about the topic? Is the aim to build new theories or hypotheses, or to establish facts and figures? Are people with learning disabilities needed as participants? Their participation should not be tokenistic but should be warranted by the research question. Some questions may be best answered by looking at records or asking carers, but for others direct interviews or observational methods will be more appropriate. If we do include people with learning disabilities, what is the best way to ensure they can participate fully? For example, what support will they need? How will we ensure they have been able to give informed consent? How much feedback will be appropriate? These are important questions, not only in ensuring that we end up with useful results, but also in ensuring ethical standards.

It must be remembered that gathering in-depth data on people with learning disabilities who have cancer is time consuming and can be emotionally demanding, so realistic costs and resources must be carefully considered, in particular including the cost of supervision and support for the researchers.

Recruitment

Recruitment of suitable participants with learning disabilities can present a major challenge. [Lennox *et al.* \(2005\)](#) reported a range of issues, including difficulties in locating suitable participants who may live in diverse settings; carers who are already too busy and tired, and who may be reluctant to take on the additional burden of supporting the research; a suspicion felt by carers towards the research; and organisational barriers, where there are too many tiers of management between the participant and the nominated liaison person. [Lee \(1993\)](#) and [Stalker \(1998\)](#) describe the

way organisations, professionals or support workers can block access to potential participants by 'gate-keeping'.

We encountered similar problems in several studies ([Tuffrey-Wijne *et al.* 2006](#)). It was often difficult to identify people with learning disabilities who had (or were affected by) cancer. In practice, the most effective method of recruitment was through personal contact (usually at conferences or teaching sessions with concerned staff or carers).

The ethical safeguards around the project prevented us from approaching potential participants directly, so some people did not hear about the research, and were not given the option of deciding whether or not to participate.

Professionals could be understandably protective about asking a potential participant and their family or carers to take part and to involve an unknown researcher, at a difficult and worrying time. Life was complicated enough with the huge array of people already involved. We found that 'gate-keepers' tended to agree to take part if they could see some benefit for themselves or the person, with comments ranging from 'it will be nice for him to have someone to talk to' to 'it's good to have an outsider to keep an eye on things'. Our perceived professional expertise in the area of cancer, end-of-life support and bereavement often facilitated recruitment, and we had to be clear about the boundaries of our role as researchers.

Researchers would do well to take the time-consuming task of gaining access to participants into account when planning their studies. Sufficient time should be allocated to the process of finding people willing to participate, gaining the confidence of staff, explaining the study, meeting the participants and gaining consent ([Owen 2001](#)). It is also time-consuming to produce accessible information materials; to collect data from people with learning disabilities; to gain trust, allowing for engagement and disengagement ([Gilbert 2004](#)); and to understand the person's method of communication. The need for such time consuming, and therefore expensive, research needs to be fully acknowledged and justified to potential funders.

Consent

Capacity to consent

The questions of how to obtain consent, how to provide accessible study information, and how to proceed if participants lacked capacity to understand such information, are of crucial importance. We were rightly asked questions about this by the Research Ethics Committee, by potential participants' carers, and by the people with learning disabilities on the research advisory group.

Some people in our study not only lacked capacity to understand the concept of a research study, but they also did not know they had cancer. We adopted the principle of

'process consent' (Department of Health 2001), giving continuous attention to the question whether the participant seemed happy to engage with the researcher. At times, this meant cutting short a planned data collection visit.

However, one can question whether it can ever be ethical to include ill and vulnerable people in a study about cancer, if they can have no idea about the purpose of the study. Is it ethical to include them in what is arguably an intrusive study, allowing a researcher access to their lives, if they cannot give their consent? We decided that the importance of understanding the experiences of those who lacked capacity to understand the reason for the researcher's presence, and of those who were unaware of their diagnosis, justified their inclusion. In fact, it could also be argued that it is unethical to exclude people with more severe learning disabilities from studies that could provide insight into their experiences and help to shape sensitive care in the future. The Research Ethics Committee agreed, but wanted to be explicitly reassured that we would not reveal any information to the participant about their diagnosis or prognosis. Carers, too, wanted this reassurance. We produced two different versions of the study information sheet: one mentioned 'cancer', the other simply 'illness'.

We can take our questioning one step further. It can be difficult for any participants, even those without learning disabilities, to understand the true nature of qualitative research and the concept of theory building. People may agree to tell us their stories, but do they also truly consent to having theory constructed that is based on those stories? An example was a close relative of Sally Burnett, a participant in the final stages of cancer, who was adamant that Sally should never be told of her poor prognosis. The hospice staff did not question this. However, over the course of several months leading up to Sally's death, it became clear to IT that Sally struggled, maybe even suffered, with the cheerful insistence of her family and hospice staff that she was doing well and might improve, and the confusing contradictory evidence of her failing body. It may be that Sally's relative would disagree with the resulting construction of theory, based not only on her perspective but also on Sally's. Very personal stories will be put into the context of wider experiences, seen through the eyes of the research team, not of the story teller. Participants thus no longer 'own' their stories. Such concepts are hard for most people to understand; how much more difficult for people with disabilities in abstract thinking.

Anonymity

We were asking participants to share deeply personal experiences. Some of them knew their stories would be published. They seemed to find the realisation that what they had gone through could be of importance to the

outside world highly empowering. Whilst some sought reassurance that IT would keep their situation confidential ('don't put my name in that book!'), two participants (John Davies and Amanda Cresswell) were very clear that they wanted to stand up and 'own' their stories. An important feature of qualitative research with people with learning disabilities is to make their stories heard, to give voice to a previously silenced group. The need to anonymise the data was sometimes very difficult to explain. Anticipating that some participants might strongly object to – or even be offended by – having their stories anonymised, we included the possibility of foregoing anonymity in the research protocol. The possible use of people's own name was given ethical approval; John and Amanda each published their stories in their own words (Cresswell & Tuffrey-Wijne 2008; Tuffrey-Wijne & Davies 2007). The literature on ethical issues in qualitative research places a heavy emphasis on the need to protect participants from harm, typically through assuring confidentiality (Baez 2002). However, some have argued that research participants' autonomous choice to waive their right to confidentiality should be seriously considered, with careful examination of the reasons for refusing the participant such a choice, as well as the implications of allowing it (Giordano *et al.* 2007). In practice, granting a wish to be identified was not an easy decision and caused considerable debate, both with the participants and within the research team. Stepping out of anonymity would also identify people's families and carers, and the participants had to understand the implications of this. John spent many months considering, trying to understand what foregoing anonymity might mean for his family, until his family gave him the reassurance that they supported his wishes. However, whilst 'being named' has undoubtedly added poignancy and power to John's story, there were unexpected ethical complications. Aspects of the care John and his family received in the weeks leading up to and immediately after his death, after his story went to press, were so shocking that IT wrote a formal letter of complaint. This is part of the research findings, but we now realise that the decision to identify John potentially means that *everyone* involved in his situation could be identified. Is this ethically sound?

Lack of anonymity could affect the rigor of the study. Researchers may feel less free to construct theory from stories that are so clearly owned by those who told them. Accommodating participants' right to refuse anonymity could therefore undermine the ultimate research objective. Despite these possibilities, we believe that it was ethically justified to give highly vulnerable participants an opportunity to leave the legacy they so desperately wanted. Being able to do this gave their illness meaning and helped them to move on in different ways: to re-build life or to accept death. It did not seem ethical to deny this opportunity. However, this warrants further debate.

Observation of sub-optimal care

At times the participant observer witnessed sub-optimal care. This raised different ethical issues. At what point should the researcher step out of her neutral role and influence the situation? The Veronica Project protocol stated that the researcher could intervene if a (lack of) care and support seemed detrimental to the participant's well-being; and that before intervening, the researcher should seek guidance from certain members of the Research Advisory Group who had agreed to provide 'ongoing' ethical advice. Intervention by the researcher would inevitably lead to a change in the person's situation and would therefore affect the data collected. However, we deemed it unethical to observe seriously inadequate care without intervention: the welfare of the participant had to come before the needs of the researchers.

In practice, this was not always straightforward. When is care sub-optimal? There were many occasions where the researcher, who had many years' experience as a nurse in the fields of both learning disabilities and palliative care and whose expected standards of care were admittedly high, observed practice that may not have been optimal, but that did not warrant intervention. Rather, these observations were simply part of the findings. Examples include participants being patronised or ignored by hospital staff, or participants' wishes not being acted on by support workers. There were a few occasions when, having consulted her Advisory Group, IT intervened. One participant, Nick Ballard, who had no speech, clearly and consistently indicated that he was in pain, but his home manager did not want to give him regular analgesia. IT's suggestions to get a palliative care review were not followed up. She became concerned when Nick began to lose his balance, a possible sign of spinal cord compression which could lead to irreversible paralysis and can be prevented by prompt treatment. After asking advice from the palliative care consultant on her Advisory Group, she spoke of her concerns to the home manager and to Nick's hospital medical team.

An observation of abusive behaviour was more challenging. Ursula Smith was very frail when a support worker at her residential care home forcibly poured coffee down her throat, despite Ursula's cries of protest and efforts to push the cup away. IT could not report this immediately, as the home manager was absent. She consulted her advisory group and reported the incident to the home manager some days later, insisting on a report to Social Services under the 'Protection of Vulnerable Adults' structure. The support worker was suspended, the incident was investigated, and this contributed to the support worker's decision to leave her employment. On reflection, IT felt that she should have intervened immediately, but such decisions can be difficult, particularly as a researcher's role is usually explicitly *not* to intervene. In new research protocols, the authors now include the clause

that the researcher may intervene without consulting her Advisory Group if he/she believes that the participant is at immediate risk from harm, working on the principle that it is unethical not to intervene when observing harmful situations.

The researcher-participant relationship

Research participants with learning disabilities are unlikely to be familiar with the role of the researcher, and this can be of particular significance in ethnographic research, where the risk of intrusion is great. Stalker (1998) highlights the concern that the researcher will be unable to meet expectations of continuing friendship. Angrosino (2004) warns that the engagement of the participant observer may be especially delicate as people with learning disabilities may have limited experience with friendships, and may have emotional needs for which the researcher is not fully prepared. Like Booth & Booth (1994) in their study of parents who have learning disabilities, IT has treated rapport as a two-way process, and has been happy to give out her home telephone number to participants and their relatives and carers (which people have used but never abused to date). She has also tried to be as clear and honest as possible about her involvement and its limitations, and to keep appointments and promises. The initial research protocol for the Veronica Project stipulated an involvement of around 4 months. However, as the study progressed, that seemed not only too short to collect all the relevant data; it also seemed unethical to withdraw what had become, in many cases, a supportive presence. While we were always aware that the researcher was not a therapist or indeed a friend in the usual sense, with some people it was clear that the researcher could not suddenly end the relationship. Booth & Booth (1994) state that 'social researchers using biographical methods with vulnerable groups must be prepared to live up to this commitment or risk their field relationship becoming exploitative' (p. 419).

In in-depth qualitative research involving people who are highly vulnerable, it is important to consider the effect of the researcher as a person on the participants. The researcher should be conscious of his/her own issues and needs, and separate them from those of the participants; otherwise, there is a real possibility of harm, not only for the participants but also for the researcher. Organising personal emotional supervision for the researcher is therefore part of doing ethical research with people with learning disabilities who have cancer or who may be dying. Although supervision by an experienced researcher and a psychotherapist was available from the start, IT had initially underestimated the extent of the need for such support to cope with the considerable emotional demands of the Veronica Project. As researchers, it is often our role to stay with people's stories and reflect on them deeply. This can sometimes be too much to bear alone. IT experienced painful tensions between her previous role as a nurse and her current role as a researcher, including the

obvious difficulty of not being in a position to take immediate practical action. Such issues must be acknowledged and talked through. It is crucial that effective structures for high quality supervision are in place and budgeted for from the start, whether or not the researcher recognises the need for such support.

Conclusion

The area of cancer and palliative care provision for people with learning disabilities is complex and multi-faceted. People with learning disabilities themselves have indicated that they want to be included and listened to, and this is one of our greatest challenges. Wherever it is methodologically appropriate they should be included as participants.

The vulnerability of people with learning disabilities who have cancer or who are dying means that such research must be conducted with the highest ethical standards. Obtaining approval from the relevant Research Ethics Committee is only a first step. There should be ongoing concern for ethical issues, with clear structures for supervision and ethical advice for the researchers. We would strongly recommend the appointment of a research advisory group, whose responsibilities include the safeguarding of ethical standards. Such an advisory group should include people with learning disabilities, whose help is also needed in developing accessible study information materials.

With careful thought and planning, and a sensitive approach, we should be able to include more people with learning disabilities in future research around cancer, death and dying; and this will benefit both participants and other people with learning disabilities.

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