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A systematic review of the experiences of vulnerable people participating in research on sensitive topics

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ABSTRACT

Objective

The aim of this article is to systematically review studies that discuss the experiences of vulnerable populations participating in research on sensitive topics.

Design

Systematic review performed according to PRISMA guidelines.

Data Sources

Thirteen databases were searched, locating 197 articles. Following removal of duplicates, screening and full text review, 31 studies remained to be critically appraised.

Review Methods

As there was a mix of qualitative and quantitative articles, the Critical Appraisal Skills Program (CASP) toolkit and Effective Public Health Practice Project (EPHPP) tool were used to appraise the methodological quality of the articles. Following critical appraisal, the remaining 11 articles were synthesised narratively to identify common themes across the studies.

Results

Despite some reports of distress, responses from participants were overwhelmingly positive. There was a strong link between symptomatology and potential for distress; however, the majority of those who did experience some level of discomfort stated they would still participate in future research. Three major themes were extracted: "It was worth it"; "Even if it hurt, I would do it again" and "Risk or benefit: fixing the location on the continuum".

Conclusion

Although researchers frequently experience obstacles and the phenomenon known as "gatekeeping" when attempting to conduct research amongst vulnerable populations, there is little evidence of harm to participants. On the contrary, there is evidence of benefit for participants and evidence that they are willing to participate if given the opportunity. Although

well-meaning, the actions of gatekeepers are not only paternalistic, they could be further marginalising vulnerable populations by denying them the benefits to be gained from research designed to identify and begin addressing their needs.

Keywords

Experience, gatekeeper, participate, research, sensitive, vulnerable

CONTRIBUTION OF THE PAPER

‘What is already known about the topic?’

- Gaining ethics approval to conduct research on sensitive topics with populations considered to be vulnerable is often a challenging process.
- If appropriate treatment is to be designed for specific groups of people, research needs to be conducted to identify those needs.

‘What this paper adds’

- Evidence of benefits from participating in research significantly outweighs the potential for harm.
- In those instances where harm occurs, it is typically not long-lasting or severe and the majority of participants are pleased they have participated and would do so again.
- Identification of strategies that can be adopted to safeguard the wellbeing of vulnerable populations participating in research.

INTRODUCTION

The need to protect participants engaged in research is inarguable, as is the obligation on researchers to uphold that protection and adhere to ethical principles of research. Without these principles, any participant is at risk of being exploited. It is similarly well recognised that some groups may require a heightened level of monitoring during research participation because they may be at increased risk of experiencing adverse reactions as a result of participating in research (Sharkey et al., 2011). Often referred to as “vulnerable” these groups can include children, the elderly, people with physical or cognitive/intellectual impairment, people experiencing serious physical or mental unwellness (particularly those with a terminal illness), people identifying as LGBTI, people taking illicit substances, the homeless, prison inmates, migrants, refugees, people from ethnic minorities, or any individuals considered to be stigmatised or marginalised (Allen, 2002). In Australia, groups

considered to be vulnerable also includes people of Aboriginal or Torres Strait Islander descent. Some have even suggested that being female defines an individual as vulnerable, while others have suggested that participants in any research study are vulnerable to some degree (Horowitz et al., 2002; Ulrich et al., 2002). Similarly, there are research topics that are considered to be “sensitive”. Asking any people about their experiences with these sensitive topics is considered to render the participants vulnerable. These topics include: bereavement (particularly bereaved parents of young children), criminal activity, serious ill health or any activity or affliction considered to be atypical.

Not only does the adoption of such broad categories of vulnerability and sensitivity have the potential to include the majority of any population, it also problematises the construction of definitions for these terms. As a result, a necessarily broad definition of vulnerability is provided, based on a description of vulnerability provided by Peternelj-Taylor (2005). Vulnerability is defined as a complex, multi-dimensional concept continually evolving according to societal values and beliefs. It typically includes people considered to be in poor health, impoverished, disenfranchised or subjected to discrimination, intolerance, subordination and stigma, or exhibiting any attributes that have the potential to result in exploitation of participants.

There is no argument that vulnerable populations may require even more stringent safeguards when participating in research; however, this vulnerability should not prevent research. Kipnis (2001) argued this point effectively when he stated that vulnerability should not be seen as a “flashing red light ordering researchers to stop, but rather as a cautionary signal, calling for proper safeguards” (p. G-4). However, there are reported instances of some ethics committees taking their protective role to the extreme that, instead of extra protection being afforded, research projects are denied ethics approval because the committee has decided that the population is vulnerable (Biddle et al., 2013). Although well meaning, such paternalism is often based on stereotyping and unfounded assumptions (Bracken-Roche et al., 2016) which may only serve to further stigmatise, devalue and marginalise groups and individuals already isolated for whatever reason. Indeed, Dennis (1999) identified this possibility when she stated:

“Neither being on the periphery of the mainstream of society nor one’s group membership should reduce one’s worth. In fact for these citizens, we must do more – not less – because when the dignity and the humanity of the most vulnerable groups is assured, it speaks eloquently of protection for all groups in society” (p. 287).

This phenomenon, more commonly known as “gatekeeping”, was defined by Walker and Read (2011, p. 14) as: “parties with an interest in ensuring that ethical standards are upheld and with some degree of influence over the granting of access to the potential study population”. Somewhat ironically, the paternalism exercised by these gatekeepers overrides autonomy, which is one of the ethical principles guiding decision making by researchers and ethics committees. Ross and Cornbleet (2003) highlighted the incongruity in such paternalistic decisions when they stated that participants were generally more willing to participate in research than their health care professionals would have expected. Similarly, Gysels et al. (2008) argued that participants were generally capable of deciding whether to participate and researchers should be mindful of their autonomy in doing so. The well-meaning but misplaced paternalism of gatekeepers also raises the very real possibility that the needs of vulnerable populations are not being met because they are not being identified. Research that could provide valuable knowledge about health and wellbeing may not be conducted because an ethics committee has decided the participants are vulnerable. Bracken-Roche et al. (2016) identified the possibility of the development of psychiatric treatment being hindered because ethics committees considered people with mental illness to be vulnerable and therefore requiring protection from research. It is often the case that care for marginalised individuals is extrapolated from care provided to other groups. However, care is not always generalisable across groups, and suboptimal care may result if treatment decisions are based on responses in other groups (Bracken-Roche et al., 2016). In order to provide appropriate care for different groups of people, individuals from those groups should be researched in order to determine their needs and design appropriate care (Beattie & VandenBosch, 2007; Kars et al., 2016). As far back as 1998, the UK government recognised the need to assess the needs of vulnerable young people (Allen, 2002). Yet, even two decades later, it still remains difficult to conduct research among vulnerable populations to the extent that some researchers give up trying or instead seek research opportunities less difficult (Allen 2002).

As shown by the results of our review, it remains the case that there is a wealth of evidence supporting the argument that many vulnerable people are willing to discuss sensitive topics, often welcoming the opportunity to talk about topics that nobody else is willing to discuss with them. They cite a range of benefits, including, catharsis, new knowledge, altruism and a new perspective or meaning to the event or experience about which they are being interviewed. Often, participants express feelings of relief that they have finally been able to tell their story to an interested listener. Conversely, there is minimal evidence of any harm (Barnett, 2001; Rivlin et al., 2012). Many of those people who do experience distress or other negative outcomes are still able to identify benefit and frequently state they were

pleased to have participated and would willingly do so again (Barnett, 2001; Biddle et al., 2013; Boothroyd, 2000). Spatz Widom and Czaja (2005) suggested that "... research studies asking sensitive and intrusive questions to potentially vulnerable individuals are not necessarily harmful, as possible risks may be offset or perceived as worthwhile by other aspects of the research experience" (p. 134). Biddle et al. (2013) extended the argument even further when they identified participant benefits from research and concluded that "Overprotective gate-keeping could prevent some individuals from gaining these benefits" (p. 356). Like so much of the complexity, diversity and multiplicity that characterises the social lives of humans, there is a need for balance in protecting individuals from harm or exploitation while simultaneously building the body of knowledge that will enhance the health and wellbeing of participants in research and the wider community.

METHODS

The purpose of this review was to systematically review the literature to examine the experiences of vulnerable populations participating in research on sensitive topics. The authors met on a regular basis to discuss the scope of the project and design the research question and methods to progress the review. From this scoping review, the research question designed to guide the full systematic review was: "what are the experiences of vulnerable participants engaged in research?" The Preferred Reporting Items for Systematic Review and Meta-analysis statement was adopted to guide the review because of its ability to guide transparent, consistent and complete reporting of systematic reviews (Moher et al., 2009).

Search strategy

As the research question was considered to be cross disciplinary, a wide range of databases (n=13) were searched, using broad search terms designed with combinations of the following words: vulnerable, marginal, stigma, participants, people, population, patients, groups, gatekeep* Databases searched were: CINAHL, Medline, Embase, SocIndex, Scopus, Science Direct, Nursing & Allied Health Database, Social Science Database, PubMed, Cochrane, Psychinfo, ERIC and Education Research.

Eligibility criteria

Articles eligible for inclusion were original research articles examining the experiences of vulnerable people participating in research. To avoid reviewer bias, it was agreed that only articles specifically describing participants as "vulnerable", "marginalised" or "stigmatised"

would be included. Review articles and conference papers were excluded. The time period for searching was extended to 1945 as that is the period when ethics in research became more formalised following the atrocities that occurred during World War II. Revelation of these atrocities eventually culminated in the Nuremberg Code in 1948, which reinforced that participation in research must be voluntary and the benefits must outweigh the risks (Mandal, et al., 2011). Only English language publications were included for review. There was no age range; however, it was acknowledged that there would be minimal (if any) research among children considered to be vulnerable. Consideration was also given to the argument that all children should be considered vulnerable (Morrow & Richards, 1996). In any event, none of the articles returned by our searches presented the results of research among children. “Grey” literature was excluded from the study because of the difficulties in assessing quality.

Article selection and screening

The initial search produced 197 records, 137 of which were duplicates. A further 51 articles were located after manually searching reference lists, resulting in 111 articles to be screened. These articles were divided between the three authors who screened title and abstract to determine match with eligibility criteria. This process identified a further 59 articles for exclusion. The remaining 52 articles were divided among all authors for full text review. The authors met on a regular basis to review progress, decisions made and also to collaborate in instances where a decision to include or exclude may not have been clear. During this process, a further 21 articles were identified that did not meet the eligibility criteria, either because they were not original research or because they reported on research design or other aspects of research rather than focusing on the experiences of vulnerable participants. Articles not considered eligible for inclusion in the systematic review were still reviewed for information that could support the narrative in both the background and discussion sections of this paper. The search and selection processes are illustrated in Figure 1.

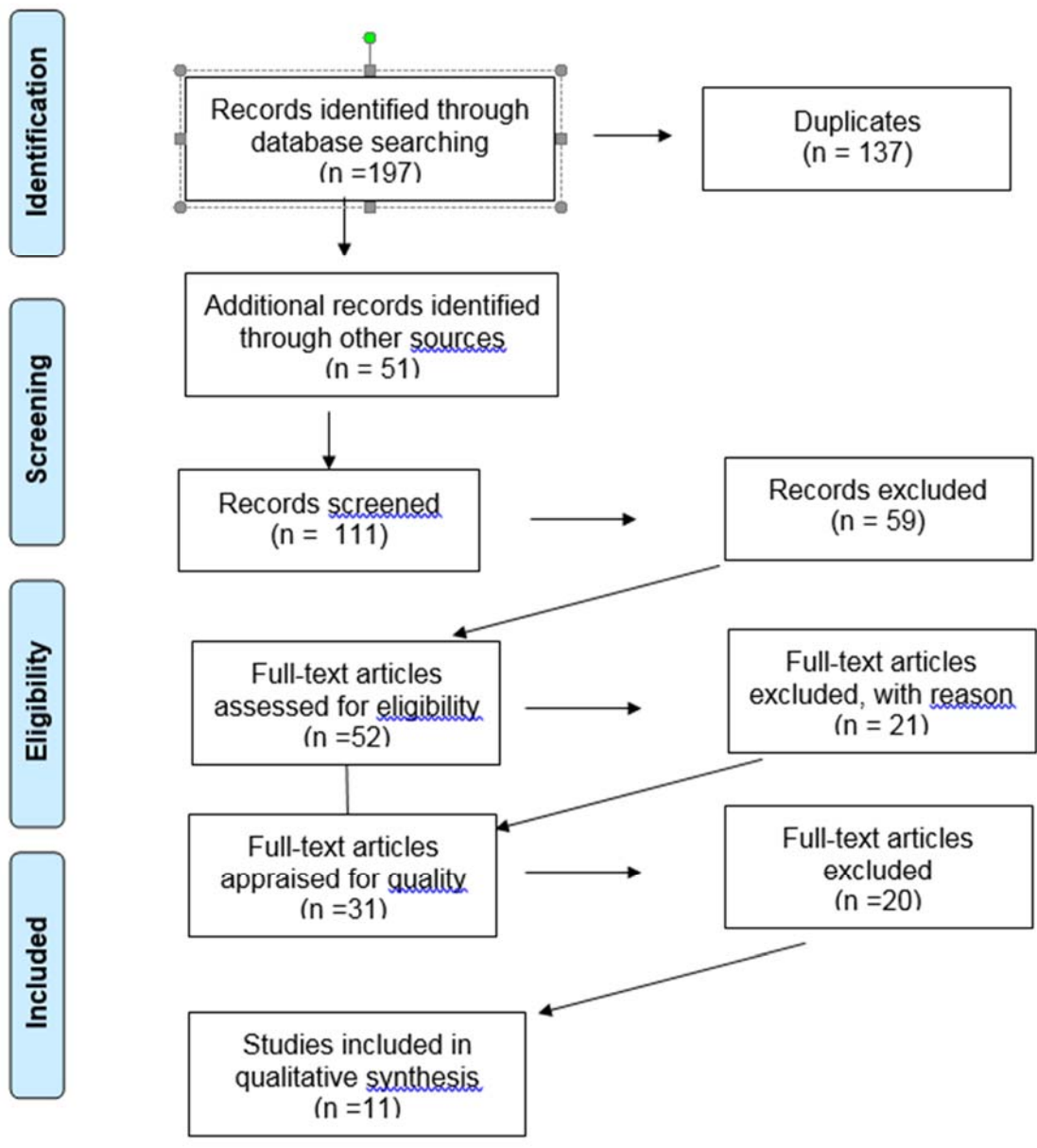


Figure 1: PRISMA flow diagram

Quality assessment

The remaining 31 articles were subjected to critical appraisal to determine their quality. As recommended by Singh (2013), standardised checklists were used to assess quality because they assist in identifying bias and other methodological weaknesses in studies,

thereby enhancing the quality of the review. All three authors conducted quality assessments. Qualitative articles were appraised according to the Critical Skills Appraisal Program (Critical Appraisal Skills Programme (CASP), 2018) toolkit, while the Effective Public Health Practice Project (Effective Public Health Practice Project (EPHPP), 1988) tool was used to appraise quantitative articles. CASP is a validated and widely used appraisal program (Singh, 2013). We would have liked to adopt CASP for the quantitative articles also, but a generic CASP quantitative appraisal tool is not available, and not all of our articles to be reviewed matched the CASP tools that were available. Accordingly, after reviewing a number of alternatives, the Effective Public Health Practice Project (EPHPP, 1998) quality assessment tool for quantitative studies was adopted. EPHPP has a strong methodological rating and has been evaluated for content and initial construct validity and inter-rater reliability (National Collaborating Centre for Methods and Tools, 2017; Thomas et al., 2004).

The critical appraisal process excluded 20 articles. Reasons for exclusion at this stage primarily related to inadequate information to support conclusions, lack of clarity, or a focus on the experiences of health care professionals. The 11 remaining articles comprised nine quantitative articles, one qualitative article and one mixed method (both qualitative and quantitative). Of these remaining articles, three rated as strong and six rated as moderate according to EPHPP. The qualitative article rated 9/10 according to CASP, while the mixed method study scored as strong (EPHPP) and 9/10 (CASP). None of the included qualitative studies provided clear affirmation about the researchers' actions in identifying the relationship between researcher and participants. This omission accounted for the less than 100% rating. Deficiencies in the included quantitative studies rating as moderate typically related to the less than comprehensive descriptions of methods adopted for the study. However, following revision of these articles, the decision was reached unanimously that there was sufficient information to indicate a robust and rigorous study, but that information was not always provided as clearly and abundantly as it might have been. Indeed, the authors noted the high number of instances where information related to a study could have been provided in greater detail which, in turn, would have improved the quality of the study. This lack of quality is unfortunate because the information contained in these articles, particularly the anecdotal comments of participants, can be a valuable descriptor of their experiences, but methodological weaknesses render the findings less reliable. Table 1 provides details of the articles included for synthesis.

Table 1: Reviewed Articles

Author, date	Design/Aims	Participants	Recruitment locations	Data collection	Data analysis	Themes/key findings	Quality score
Boothroyd, 2000	Survey to investigate the impact of participating in research	523 adults with severe mental illness	Florida (USA) care facility and following discharge to community	Structured interviews, questionnaires (Likert scale) and surveys, including: Brief Symptom Inventory, Lehman's Quality of Life Scale; MHSIP Satisfaction	Inferential statistical analysis reporting odds ratios and correlations	<ul style="list-style-type: none"> • 96% reported positive experience. • 8.8% reported experiencing anxiety. • 86% would participate again. • Participants felt valued, important, hopeful of change. 	Strong (EPHPP)
Carlson et al., 2008	Survey to assess distress and/or usefulness of participating in research interviews	206 adults with PTSD and childhood incident of physical or sexual abuse	Psychiatric inpatients	Questionnaire plus a range of psychiatric assessment tools: Structure Interview for PTSD; Physical Violence scale of the Conflict Tactics Scales; Dissociative Experiences Scale; Symptom Checklist-90-Revised; Structured Interview for Self Destructiveness	Inferential statistical analysis reporting correlations. Descriptive analysis of content	<ul style="list-style-type: none"> • 70% reported low to moderate levels of distress. • 51% found it useful. • Benefits: new perspectives, chance to help others (altruism), catharsis, some good memories too. • Negatives: reminders, embarrassment, shame. 	Strong (EPHPP)
Emanuel et al., 2004	Prospective cohort study to	Terminally ill persons	5 metropolitan and 1 rural site	Questionnaire developed to	Descriptive analysis via	<ul style="list-style-type: none"> • 89% of patients and 90% of 	Moderate (EPHPP)

	<p>assess whether interviewing people about death, dying and bereavement is stressful and/or helpful</p>	<p>(n=988) and their caregivers (n=893)</p>	<p>in USA. Random selection of physicians to identify patients. Participating patients nominated caregivers.</p>	<p>examine symptoms; social supports; communication with health care providers; spiritual and personal meaning of dying; care needs; end-of-life plans; economic burdens; sociodemographic characteristics</p>	<p>variance for age; analysis of education and income. Inferential analysis via tests of independence; bivariate analyses; stepwise logistic regression</p>	<p>carers reported little or no stress.</p> <ul style="list-style-type: none"> • 7% of patients and 8.4% of carers reported some stress. • Even those reporting stress participated in the second interview. • 47% of patients and 53.5% of carers found the research to be helpful. • 50% of patients and 45% of carers did not find the research helpful. 	
<p>Griffin et al., 2003</p>	<p>To examine reactions to trauma assessment procedures in physical or sexual assault</p>	<p>170 survivors of interpersonal violence</p>	<p>Domestic violence shelters and victim assistance agencies</p>	<p>Self-report questionnaires plus psychophysiological assessment including: heart rate and skin conductance. Psychological assessment via Clinician-Administered PTSD Scale and the Structured Clinical Interview for DSM-III-R</p>	<p>Descriptive statistical analysis, citing mean ratings and standard deviations</p>	<ul style="list-style-type: none"> • 5% would not participate again. • Very positive experience for the majority. • Participants reported participation to valuable. • Distress higher in the context of PTSD (greater symptomatology). 	<p>Moderate (EPHPP)</p>

Gysels et al., 2008	To explore patients' and carers' preferences and expectations regarding their contribution to research in palliative care	Patients (n=79) and carers (n=25) receiving palliative care	London (UK) teaching hospital	Semi-structured, open-ended exploratory interviews	Thematic analysis	<ul style="list-style-type: none"> • Not allowing vulnerable people to participate is not "protecting" them. It is denying their autonomy. • Motivation to participate related to: altruism, gratitude, concerns about care, need to talk to somebody, need for information or services. • Participants were capable of deciding for themselves and negotiating their participation. 	9/10 (CASP)
Halek et al., 2005	To assess the impact of a potentially distressing mailed survey on the emotional wellbeing and health care	3,337 veterans experiencing PTSD	Questionnaires mailed to homes of veterans seeking compensation for service-related PTSD	Questionnaires	Descriptive statistical analysis, citing mean ratings and standard deviations	<ul style="list-style-type: none"> • Distress rare (2.7%), but even 13% of those reporting distress still verbalised positive outcomes. • Health care utilisation (HCU) 	Strong (EPHPP)

	utilisation of US veterans					<p>decreased following research survey.</p> <ul style="list-style-type: none"> • Participants expressed gratitude that somebody was interested and listening to their stories. • Reduced suicidal ideation. • Increased symptomatology (particularly PTSD) linked to increased chance of distress, but still rare. 	
Parslow et al., 2000	To examine the potential for epidemiological studies, specifically related to PTSD, to cause further harm to participants	641 Australian Army Vietnam veterans experiencing PTSD	Random mailout of invitation to participate	A variety of mental and social health measures adopted from the Structured Clinical Interview for DSM-III-R; AUCSID-V;	Inferential statistical analysis via hierarchical logistic regression. Descriptive statistical analysis, citing mean ratings and standard deviations	<ul style="list-style-type: none"> • 75% with current PTSD (56.5% with past PTSD and 20.6% with no PTSD) experienced distress associated with participation, but only short-term and still willing to participate again, and no increase in HCU following participation. 	Moderate (EPHPP)

						<ul style="list-style-type: none"> Higher symptomatology linked with greater potential for distress. 	
Pessin et al., 2008	To assess the burden and benefits of participating in psychosocial research addressing end of life issues of persons receiving palliative care	68 terminally ill persons	Patients admitted for end of life care at a New York hospital	Interview plus clinician-administered and self-reported assessment tools, including: Benefit and Burden Scale	Descriptive statistical analysis via frequencies. Inferential statistical analysis reporting correlations	<ul style="list-style-type: none"> 75% reported no burden from participating. 68% reported participation to be moderate to highly beneficial. 6% reported a high level of distress. 77% agreed they would participate again. 	Moderate (EPHPP)
Rivlin et al., 2012	Case control study to investigate the effects of participating in detailed interviews about suicidal behaviour	120 prison inmates post suicide attempt compared to 120 prison inmates who had not attempted suicide	19 male and 10 female prisons in the UK with high rates of completed or attempted suicide	Interview plus self-reported mood assessment using a Visual Analogue Scale. Assessment tools: Oxford Monitoring System for Attempted Suicide; Mini International Neuropsychiatric Interview; Life Events and Prison Experiences Questionnaire; Childhood Trauma Questionnaire;	Descriptive statistical analysis of quantitative data. Thematic and content analysis of qualitative data.	<ul style="list-style-type: none"> Some participants reported being upset, but majority pleased to participate. Symptoms of distress reduced after participation. 75% reported improved mood after participating. 2% reported deteriorated 	Strong (EPHPP); 9/10 (CASP)

				Social Support Scale		<p>mood after participating.</p> <ul style="list-style-type: none"> • Benefits: enjoyment, altruism, new perspectives, catharsis. • Higher symptomatology linked with greater potential for distress. 	
Scott et al., 2002	Retrospective study to investigate participants' experience of involvement in a study following their child being diagnosed with Ewing's sarcoma	81 parents of children with Ewing's sarcoma in Queensland, Australia	Participants in a previous study were invited to participate in a study investigating their research experience	Mailed self-administered follow up questionnaire containing open and closed questions	Descriptive statistical analysis, comparing questionnaire responses. Inferential statistical analysis, citing correlations and probabilities	<ul style="list-style-type: none"> • 97.5% glad to have participated and hoped it would benefit others. • Participation more distressing for those whose child is still alive, than for those whose child had died. • Participation painful, but would do it again. • None disagreed with the statement that they were not glad to have participated. 	Moderate (EPHPP)

						<ul style="list-style-type: none"> • Benefits: chance to talk and new perspectives. 	
Spatz Widom & Czaja, 2005	To investigate the extent to which vulnerable individuals react to participation in research, compared to non-vulnerable individuals in an effort to support evidence based decisions about the participation of vulnerable people in research	896 individuals deemed to be vulnerable as defined by economic, psychological, social, physical health or child maltreatment status	Adult participants in a longitudinal study originally designed to investigate the long-term consequences of maltreatment and neglect	Participants were asked 8 questions following their participation in the longitudinal study. The questions were adapted from the Reactions to Research Participation Questionnaire.	Descriptive statistical analysis, citing prevalence, range and standard deviation	<ul style="list-style-type: none"> • Increased vulnerability linked with increased reaction when compared to non-vulnerable, but no difference in willingness to participate. • Found the research to be meaningful. 	Moderate (EPHPP)

Data extraction and synthesis

In view of the fact that both qualitative and quantitative studies were to be included in the review, the decision was made to synthesise the findings narratively in line with the guidance provided by Popay et al. (2006). Narrative synthesis is an approach that relies on the use of text and words to summarise the findings of a research study. The aim is to “tell the story” of the findings from the included studies, whether they are qualitative, quantitative or mixed method (Popay et al., 2006). It is an ideal method to compare findings and extract themes from heterogeneous studies.

The first step was to develop a framework based on the authors’ anticipations of what the data might find. This step was useful because it facilitated the identification of the authors’ expectation that the data would highlight a significant imbalance between the positive and negative consequences for vulnerable people participating in research. Having identified this bias, the authors were careful to ensure that negative instances were fully reviewed and included in the findings. It would have been helpful to be able to state that we had fully obliterated this bias from our review. However, as identified by many researchers, it is not possible to fully remove the influences of our biases (Denzin & Lincoln, 2017; Nagel, 1986). The best we can hope for is to recognise them and reduce their influence as much as possible.

The second step focused on developing a preliminary synthesis. Two of the authors worked together to review each of the 11 articles in detail and extract the findings and other information that could inform our research question. At this stage, it was already becoming apparent that our anticipation of a significant imbalance was likely to be correct.

In the third stage, all three authors met to compare the findings extracted from the articles, to explore relationships and identify patterns within the data. The multiple pieces of evidence from the individual studies were combined to construct a concept map that facilitated the identification of a number of sub themes, while clearly illustrating the three main themes that have become the basis for the current article.

Findings

Despite extending the search period to 1945, the earliest article to be reviewed was dated 2000. We suggest two possible reasons for the lack of earlier articles. Firstly, although currently still a difficult process with many obstacles to overcome, researchers wishing to conduct research with people considered to be vulnerable faced even more barriers in the

past, to the extent that research projects did not proceed because of the perception that vulnerable participants would be harmed. Such restrictions may have been well meaning and related to a desire to protect participants; however, as noted elsewhere, this approach could be considered paternalistic and not only overriding participants' right to autonomous decision making, but also denying them and the wider community the benefits to be gained from research. There is no argument that participants must be protected, but that "protection" must be balanced and not progress to making decisions for them. The second postulated reason for the lack of earlier articles is that the quality of publications has increased markedly in the last two decades and the methodologies described in earlier articles may not have rated adequately for inclusion in this review.

Analysis through narrative synthesis facilitated the extraction of three main themes and five subthemes. The first two themes are presented from the perspectives of the participants as reported in the reviewed articles, whereas the final theme is presented from the perspective of the authors of the articles as they presented their central arguments. Although there is significant overlap and reciprocity between the themes and subthemes, they have been delineated as far as is possible to illustrate the findings of this review. The themes and subthemes are illustrated in Figure 2 and expanded upon in the ensuing findings and discussion sections of this paper.

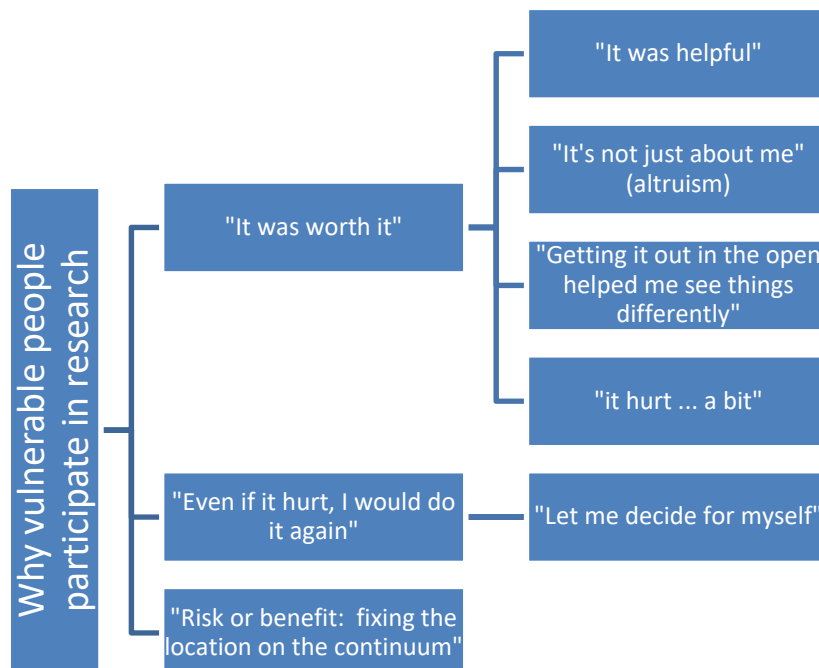


Figure 2: Themes and sub-themes

Theme 1: "It was worth it"

Although not every participant reported the experience to be helpful, the vast majority agreed that any negatives associated with participating in research were significantly outweighed by the positives, making the experience worthwhile overall. This interplay is perhaps best illustrated by Griffin et al. (2003) when they stated that:

"... existing empirical data also indicate that research participation does not overwhelm or retraumatize individuals and that benefits can be derived from participation even when some distress is experienced. Research participation is typically described by participants as a positive or sometimes neutral experience that they would be willing to repeat" (p. 222).

Some of the benefits identified by participants included: altruism, gratitude, somebody to talk to, an interested listener paying attention to their story, the opportunity to develop new perspectives, reduced health care utilisation, social interaction, catharsis, distraction, advocating for self and own autonomy, and enjoyment. Many participants verbalised their appreciation of the opportunity to have somebody to talk to. For some, it was their first opportunity to talk to an interested independent person and know they had been heard. Negative aspects of participating in research included experiences of distress and discomfort frequently related to the triggering of painful memories. As it is not possible to discuss all of the positive comments, a selection has been provided in Table 2.

Author	Title	Extracted comments
Boothroyd, 2000	The impact of research participation on adults with severe mental illness	96% reported the interview to be a positive experience (on a spectrum of slightly to very positive).
Carlson et al., 2008	Distress in response to and perceived usefulness of trauma research interviews	51% found the research useful.
Emmanuel et al., 2004	Talking with terminally ill patients and their caregivers about death, dying and bereavement	47% of patients and 53.5% of carers found the research to be helpful. 89% of patients and 90% of carers reported little or no stress associated with the research.
Griffin et al., 2003	Participation in trauma research: Is there evidence of harm?	Very positive experience for the majority.
Halek et al., 2005	Spontaneous reports of emotional upset and health care utilization among veterans with post traumatic stress disorder after receiving a potentially upsetting survey	Even 13% of those reporting distress still verbalised positive outcomes. Participants expressed gratitude that somebody was interested and listening to their stories. Possible the survey was actually therapeutic for some participants.
Pessin et al., 2008	Burden and benefit of psychosocial research at the end of life	68% reported participation to be moderate to highly beneficial.
Rivlin et al., 2012	Impact on prisoners of participating in research interviews related to near-lethal suicide attempts	Symptoms of distress reduced after participation. 75% reported improved mood after participating.
Scott et al., 2002	Does research into sensitive areas do harm? Experiences of research participation after a child's diagnosis with Ewing's sarcoma	97.5% glad to have participated and hoped it would benefit others.
Spatz Widom et al., 2005	Reactions to research participation in vulnerable groups	Found the research to be meaningful and felt treated with respect and dignity.

Table 2: Extracted comments from reviewed articles

Subtheme 1.1: "It was helpful"

Even though we do not intend to claim therapeutic benefit, there are indications that participants found participation to be helpful therapeutically. Boothroyd (2000), Halek et al. (2005), and Pessin et al. (2008) all suggested participants benefitted therapeutically from the research interview. Rivlin et al. (2012) produced evidence to support their claims of therapeutic benefit from the interviews and stated that participation "... can be beneficial" (p. 54). Emanuel et al. (2004) suggested that: "... having the discussion and expressing interest may be therapeutic" (p. 2004).

Halek et al. (2005) were able to provide actual evidence of therapeutic benefit from their study. They were able to identify two participants who were actively suicidal during the study's progress. The authors reported they were able to provide rapid assistance to these "otherwise isolated" individuals who had not known whom to call for assistance. The issue of social isolation was also mentioned by Gysels et al. (2008). They found that participants with mobility restrictions welcomed the opportunity for social interaction presented by the research interview, viewing it as a "release" from their daily isolation.

Given the positive impact and indications of therapeutic benefit presented in the reviewed articles, it is reasonable to assume a flow on effect of decreased utilisation of health care services. However, Halek et al. (2005) were the only authors to specifically include this indicator in their research. Parslow et al. (2000) did consider health service utilisation following participation in a research interview, but their interest was in whether the interview itself resulted in increased access of services, rather than investigating any possible therapeutic benefit.

"After they received a potentially distressing questionnaire, our subjects' mental health care utilization and total health care utilization decreased significantly" (Halek et al., 2005, p. 149).

The authors hypothesised that the act of writing about a previously undisclosed event may have contributed to feelings of release and relief, and that this heightened sense of wellbeing resulted in decreased need to access health care.

Although not specifically linking to reduced health care utilisation, it is not unreasonable to interpolate the findings of Rivlin et al. (2012) of improved mood among prisoners to a subsequent reduced need for health care services: "... self-reported mood levels increased significantly by the end of the interview compared with at the beginning" (p. 61).

In view of the fact that there is little empirical evidence of the influence of research participation on subsequent health care utilisation, this could be an area for future research.

Subtheme 1.2: "It's not just about me" (altruism)

Verbalisation of feelings of altruism are common among research participants, even those discussing sensitive topics. They often express sentiments such as "I will do it if it helps others" and "save people from what they had to go through" (Gysels et al., 2008, p. 352). Gysels et al. (2008) investigated the reasons why patients receiving palliative care chose to or declined to participate in research. They found that of the "few patients" who declined to participate, the most common reason was ill health, which would not be an unexpected response in a palliative care population. Of the reasons for participation, the authors described as "striking" their finding that all participants with motor neurone disease (MND) stated altruism as their reason for participation.

"Some explained that they had made a conscious decision to take part in any research on MND from the moment they had been given the diagnosis. They expressed their wish to contribute to anything that would raise greater awareness and knowledge about this rare disease, so that more could be done to save people from what they had to go through ... They found that an interview was the least they could do" (Gysels et al., 2008, p. 352).

Similarly, US Army veterans expressed altruism as a reason for their participation: "Most common in these situations, veterans thanked us for our interest in such important topics (e.g., sexual trauma) or expressed hope that their responses could help others" (Halek, et al., 2005, p. 146).

Even if participants did not find the research participation to be personally useful, they were willing to participate if it would help others: "... many participants comment that it was not useful to them personally but that they hoped it would help others ... (Carlson et al., 2008, p. 134).

A participant in the study by Carlson et al. (2008) expressed the hope that her participation would "... help stop this abuse" (p. 138). She hoped the project would increase understanding of the experiences of trauma survivors, even if it was only the understanding of the researchers that was increased.

Losing a child is recognised as one of the most traumatic experiences that people endure. However, even with this significant personal burden, 97.5% of participants in a study investigating the experiences of parents of children with Ewing's sarcoma stated they were glad to have participated and hoped it would benefit others. Almost half of the participants in this study believed the research interview had been an opportunity to achieve some good from an otherwise bad situation. One participant acknowledged the interview was painful, but stated the pain would not have been in vain if it saved just one child from suffering as her own daughter had (Scott et al., 2002).

Altruism was also mentioned as a reason for participation in the study by Rivlin et al. (2012). Participants commented that they liked "... giving stuff back ..." and were "... more than willing to help" (p. 57). Another participant expressed the hope that "... it might help to prevent someone from hurting themselves" (p. 58). One participant stated their willingness to help the researcher: "You wanted help with your job ... I hope I will help people and help you" (p. 58).

Due to the frequency with which it is mentioned, altruism could be considered to be one of the primary reasons people participate in research. The findings of Rivlin et al. (2012) support this suggestion: "... the belief that the research might help someone else was the most common 'best' element of participation" (p. 58).

This finding related to the control cohort of the study, but even participants in the case cohort rated altruism as the second most important for participation. Their primary reason for participating was the personal benefit they obtained through talking to an interested person about their problems.

Subtheme 1.3: "Getting it out in the open helped me see things differently"

Up to this point, we have presented findings related to the impact on health services and people generally. However, participants were also able to articulate many personal benefits of participating in research. They spoke of feelings of catharsis, gratitude, distraction and others that all contributed to them developing new and more helpful perspectives on their experiences.

Psychiatric inpatients in the study by Carlson et al. (2008) selected "Led to new insights" as being the primary measure (35.6%) of usefulness of their participation. The next most

important measure (16.4%) was the opportunity to talk to somebody else. One participant in that study stated “It put things in perspective. When I look at my life, I can understand why I was so scared” (p. 138).

Although acknowledging the lack of specific understanding about how research contributed to new perspectives, Emanuel et al. (2004) suggested that: “... answering structured questions helped them better understand their experiences, or that, for many people, having someone, even unknown, be interested in hearing about their personal experiences at this sensitive moment is helpful” (p. 2003).

Participants in the study by Rivlin et al. (2012) discussed their feelings of catharsis “... gotten it off my chest” which was better than “... having it all bottled up all the time” (p. 59). They typically agreed these processes helped them to develop new perspectives by “... thinking really deep about issues” (p. 59). Gysels et al. (2008) reported that participants expressed appreciation for the opportunity to “vent frustrations” (p. 352). Participants in this study also expressed their appreciation of the opportunity to:

“... talk with an interested outsider about the problems and uncertainties faced and the misunderstandings that had arisen with health professionals ... After the interview, patients often expressed their thanks for having been able to make sense of their experiences and said that they should have had this opportunity much earlier” (p. 352).

Participants in Pessin et al. (2008) stated that the study was “... helping to keep them busy” as well as helping them “... think about issues they had not necessarily considered or discussed ...” (p. 630).

Subtheme 1.4: “It hurt ... a bit”

Despite experiences being overwhelmingly positive, there was some evidence of distress associated with participation in research. Distress was more frequent and more intense for those with higher symptomatology and those recounting traumatic events. Even so, Griffin et al. (2003) found participation was well tolerated and survivors were not too fragile to participate even after severe trauma.

In the majority of cases, any reports of distress were relatively low, typically below 10% of participants, while reports of severe distress were typically below 5%. Some studies did report on levels of withdrawal because of discomfort, but these rates were small, generally below 5%. Reasons such as deteriorating health or relocation accounted for higher rates of withdrawal than the reason of discomfort. Triggers for distress were frequently related to painful reminders, embarrassment and shame.

The highest rates of distress were reported in the study by Parslow et al. (2000) among Australian Vietnam veterans. 75% of participants with current PTSD and 56.5% of participants with past PTSD reported distress, while only 20.6% of those not experiencing PTSD reported distress. These figures provide further evidence of the link between higher symptomatology and greater distress. However, the distress that was experienced was described as “short lived” and not resulting in increased health care utilisation. High rates of distress were also recorded in the study by Carlson et al. (2008) where 70% of participants reported “relatively low levels of distress” (p. 132). However, the authors did note that the cohort of psychiatric inpatients probably represented a “worst case scenario” (p. 140). Boothroyd (2000) also conducted research with participants experiencing mental health disorders, but only 4% of their participants described the research participation as negative. It is possible that participants in the study reported by Carlson et al. (2008) experienced these levels of greater distress because they were recounting traumatic experiences. Discussion in the next theme will expand on this finding of the current review that higher symptomatology and other burdens are positively linked to the potential for higher levels of distress associated with research participation.

Theme 2: “Even if it hurt, I would still do it again”.

Apart from the high incidence of positive experiences from participating in research, the other almost universal finding across the studies was the positive correlation between higher symptomatology and reports of distress. However, even those participants with higher symptomatology and experiencing some level of discomfort frequently stated they still found the experience helpful and were pleased they had participated. They were significantly more likely than unlikely to participate in future research projects. Despite participants in Parslow et al. (2000) recording the highest incidence of stress associated with research participation, the majority stated they would still participate again. Similarly, Spatz Widom and Czaja (2005) found that: “... psychologically vulnerable individuals more strongly agreed they would continue to participate ... and found their participation meaningful” (p. 115).

Carlson et al. (2008) highlighted the “interesting paradox” of: “... the most prominent reason given for why the interview was upsetting (remembering the past) was also the means of achieving the most prominently reported benefit (led to new insights)” (p. 139). They suggested that: “... what is upsetting about participating in trauma interviews may be inextricably entwined with what is useful about participating” (p. 140).

In the study reported by Scott et al. (2002), even though almost 50% of participants anticipated that the interview would be painful, 93.8% were pleased to be involved. Despite experiencing the death or terminal illness of a child, no participant in that study disagreed with the statement “I am glad to have participated in the interview” (p. 509). The high rates of distress experienced by participants in the study by Parslow et al. (2000) have already been presented, but most of these participants expressed willingness to participate again. The study by Emanuel et al. (2004) incorporated two interviews. Even those participants experiencing distress in the first interview were mostly still willing to participate in the second interview. Griffin et al. (2003) conducted research among survivors of sexual assault – an undoubtedly traumatic experience – but 95% of participants said they would participate again.

Subtheme 2.1: “Let me decide for myself”

Importantly, participants stated their desire to decide for themselves whether they would participate in research. Gysels et al. (2008) concluded that participants were:

“... capable of deciding whether to participate in interviews and negotiating how they wanted this to happen” (p. 347) and “It strengthens our position that research should respect patients’ autonomy and enable their voices to be heard when they choose to participate” (p. 355).

Theme 3: Risk or benefit: fixing the location on the continuum

All of the studies reviewed for this systematic review concluded that any risks for vulnerable populations associated with participating in research were significantly outweighed by the benefits. Any distress that did occur was typically minor, short-lived, and not sufficient to deter participants from participating in future studies. To avoid the risk of belabouring this point, we have not included the concluding remarks of all eleven reviewed articles, but

elected instead to present the most eloquent. For example, Gysels et al. (2008) articulated their argument clearly by stating:

“From this perspective, the extreme position of arguing that palliative care patients should never be asked to take part in research is not justified. Simply because patients are nearing the end of life is not a valid reason to exclude them from research. To do so implies that they also will be denied the benefits of participating in research studies and having their voices heard” (p. 353).

While Pessin et al. (2008) suggested that: “... conducting psychosocial research can be minimally burdensome to a palliative patient if conducted in a sensitive manner, and in fact, in some cases may be beneficial” (p. 630).

Rivlin et al. (2012) concluded that: “... it may be helpful for ethics committees to be more aware that in some cases there may be potential therapeutic benefits to be gained from research participation beyond clinical trials, even when it involves examining traumatic experiences ...” (p. 62).

Scott et al. (2002) concluded:

“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” (p. 507).

Finally, Spatz Widom and Czaja (2005) concluded that their results demonstrated:

“... that these results and similar ones from other studies indicate that researchers and IRBs should not be wary of conducting research on sensitive topics with potentially vulnerable populations, particularly research that has the potential for further understanding the characteristics or needs of these kinds of vulnerable populations” (p. 136).

We consider these statements illustrate the current state of evidence so well that further comment would be superfluous.

DISCUSSION

Supported by the foregoing evidence, we argue that any potential for harm associated with participating in well-designed research is significantly outweighed by the potential benefits. The evidence reviewed and presented in this paper supports the contention that participating in research is unlikely to cause harm to participants considered to be vulnerable, even when sensitive topics are discussed. In fact, far from avoiding harm (the minimum requirement of research), evidence has been presented that supports the possibility of participation in research having therapeutic benefit. Rivlin et al. (2012) provided an example of the complicated continuum that characterises the benefit versus harm argument. They interviewed prison inmates who had had attempted suicide and found that self-reported mood levels improved or did not deteriorate significantly for almost all prisoners. Although some prisoners found the interviews to be upsetting, nearly all were pleased that they had participated.

Evidence of research benefit discussed in this paper refers primarily to vulnerable cohorts. For example, 75% of participants in the bereavement study by Seamark et al. (2000) cited the experience as helpful or very helpful, implying some evidence of therapeutic benefit. However, it is not unreasonable to extrapolate evidence from research with populations not considered to be vulnerable. Participants in a study by Castillo et al. (2012) cited empowerment and new knowledge as benefits of their participation, while Josselson (2007) stated that most people found research interviews to be “healing, integrative, useful and meaningful” (p. 559). There is also wider evidence of altruism being a motivating factor for people deciding to participate in research. Carrera et al. (2018) argued that a “... sense of empathy and shared connection with others is a key factor in the decision to participate in research” (p. 175). Similarly, participants in a study investigating experiences of bereaved spouses cited altruism as one of the main reasons for participating (Seamark et al., 2000). Participants frequently experience feelings of satisfaction or enhanced wellbeing in their belief that they are benefitting others, even if they do experience some pain from doing so.

Experiencing catharsis was another benefit frequently described as an outcome of participating in research. Although participants in the bereavement study by Seamark et al. (2000) cited altruism as the main reason for participating, catharsis rated as the second most important reason. Women engaging in transactional sex in Florida (USA) expressed feelings of catharsis during interviews, giving particular value to the opportunity to tell their story to a non-judgemental listener. They also described feelings of altruism and heightened insights into their own emotions and behaviour (Felsher et al., 2018).

We agree that there is some risk of distress for those participating in research, particularly when sensitive topics are being investigated. However, any distress or discomfort is typically not severe or long-lasting. Sikweyiya and Jewkes (2013) examined reports of discomfort arising from participation in research interviews. They found that discomfort did not amount to psychological harm and cited other research studies that found that participants were not emotionally or psychologically harmed by talking about their traumatic experiences. Indeed, we would argue that there is a greater risk for harm to be caused by inappropriate (uninformed) care that is based on assumptions generalised from other cohorts because there is insufficient evidence upon which to design care. Taking palliative care as an example, the need to build the evidence base is recognised, but researchers continue to experience obstacles when attempting to recruit patients with terminal illness (Kars et al., 2016; Williams et al. 2006). As a result, evidence is often generalised from curative care that could be wholly inappropriate to a dying patient and their family (Author blinded, 2010). This argument was supported by Ross and Cornbleet (2003) who acknowledged the lack of a strong evidence base for pharmaceuticals and other therapies in advanced disease, primarily because of difficulties recruiting participants into the research studies that would strengthen the evidence. To continue building the evidence base for the care of people considered to be vulnerable, it is essential to continue conducting research with them.

Discussion in the background section of this paper has already identified the necessity for any research with human participants to stringently adhere to ethics standards and principles. The primary bioethics principles guiding health research are autonomy, beneficence, non-maleficence and justice (Johnstone, 2015). However, it could be argued that most, if not all, of these ethics principles are being overridden if vulnerable populations are excluded from research. While it is acknowledged that vulnerable populations are at greater risk of exploitation through their vulnerability, the responsibility is also acknowledged for researchers to embed even greater safeguards in their research projects to ensure the protection and wellbeing of participants who may be considered vulnerable. As research with vulnerable populations continues to increase, a growing number of researchers are suggesting safeguards that will uphold the wellbeing of participants, ensuring at least that the principle of non-maleficence is being met. van Wijk and Harrison (2013) provided a comprehensive list of safeguards, while Sharkey et al. (2011) highlighted the importance of explicit explanation of the content of interviews, and Rivlin et al. (2012) suggested conducting mood surveys prior to the interview. Sikweyiya and Jewkes (2013) also reviewed special precautions that would assist in ensuring appropriate protection for vulnerable groups in research. The reiteration in this review of the positive correlation between symptomatology and increased risk of distress further reinforces the need for careful design

of research investigating sensitive topics. However, this requirement does not equate with overriding the autonomy of potential participants and preventing them from deciding for themselves whether or not to participate in something that may be of benefit and, at the least, run only a small risk of causing distress. In contrast to the enduring assumptions among some ethics committees, there was no evidence in any of the papers reviewed for this study that ethics principles were violated or that any vulnerable participants were exposed to unacceptable risk.

The bioethics principles of autonomy and beneficence are of particular significance when discussing research with vulnerable populations because people considered to be vulnerable have had their right to autonomy overridden in the past and been denied the benefits to be gained from participating in research. As a number of authors have argued, these actions are themselves ethically questionable (Adderley & Smith, 2007; Krouse et al., 2003). Instead of questioning whether people considered to be vulnerable should be involved in research, a more appropriate question might be to ask if it is ethical to exclude them (Bradburn & Maher, 2005). Excluding marginalised people from research exposes them to the risk of even greater marginalisation, thus overriding the bioethics principle of justice. Some might argue that the bioethics principle of non-maleficence is being upheld by paternally deciding not to recruit vulnerable participants. However, if there is minimal or no risk and if safeguards are employed to reduce even further the slight chance of harm occurring, that should satisfy the requirements of non-maleficence without the need for a totalitarian and exclusionary approach that subsequently overrides other ethics considerations.

CONCLUSION

The territory of research with vulnerable cohorts is complex. As is the case with any complex situations, there are no simple answers. However, there are some nuggets that can guide the conduct of research with vulnerable populations. All research participants must be protected from harm, particularly when sensitive topics are being investigated. It is imperative that these safeguards are embedded in research design, with particular attention to the likelihood of greater symptomatology resulting in greater distress. However, it must be remembered that, for the vast majority of participants, research is neutral or may even provide benefits such as altruism, catharsis, new knowledge or new perspectives. Paternalistic decisions not to allow research with vulnerable individuals not only overrides

their autonomy and denies them the benefits for themselves and the wider community of participating in research, it also overlooks or devalues their own personal agency. In the limited number of cases where discomfort or distress is recorded, it is typically short lived and not reaching the level of psychological or emotional harm. Finally, even when vulnerable participants do experience distress, the majority are glad that they participated and would do so again. These factors all support the argument that the risk/benefit ratio for research among vulnerable populations is firmly skewed to the benefit end of the continuum.

Limitations of the review

We are aware there are a number of reports presenting the results of research amongst vulnerable populations. These reports may also have included valuable anecdotal and statistical information about the experience for participants. However, as these studies were not focusing on the experience of participation, which was the main focus of this project, they have not been included for review. Another possible limitation is our decision to only review articles describing their participants as “vulnerable”, “marginalised” or “stigmatised”. This decision was made to avoid any bias that may have resulted if the reviewers decided which cohorts were or were not vulnerable.

Recommendations for research

Although there is scope for further research into the experiences of vulnerable individuals participating in research generally, there are some other areas identified by this review that are worthy of further investigation. Firstly, as highlighted by Sikweyiya and Jewkes (2013), the agency of participants and ability to protect themselves from harm has received minimal attention previously and is currently at risk of being overlooked. While it would be dangerous to assume that every participant had the same level of agency, or even that there is a standard power ratio in the researcher/participant relationship, overlooking an individual's ability to protect themselves from harm is once again entering the territory of paternalism. Secondly, as highlighted by Halek et al. (2005), it would also be useful to investigate participants' utilisation of health care services following participation in research on sensitive topics. This research could be dual purpose: to identify whether the level of distress associated with research was sufficient to lead to an increase in health care utilisation; or, to investigate whether the participant benefitted from research participation to the extent that they reduced their utilisation of health care. Thirdly, as discussed by Sharkey et al. (2011), the option of collecting data online is another option worth considering. Although it has its

own challenges (confidentiality, verification, security), there are benefits (anonymity, perceived safety) that may make it a viable alternative, particularly when researching sensitive topics.

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