

THE UNIVERSITY OF HULL

Cancer Screening, Diagnosis, and Treatment Experiences for People with Learning
Disabilities: Staff and Patient Perspectives

being a Thesis submitted in partial fulfilment of the requirements for the degree of

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Overview

This portfolio thesis has three separate parts: a systematic literature review, an empirical paper, and corresponding appendices.

Part One is a systematic literature review that aimed to identify the factors affecting the psychosocial experience of cancer for people with learning disabilities. Eight papers were identified by the database search, the findings of which are reviewed in a thematic synthesis. Four themes were derived from the data: Communication, Feeling Dismissed, Physical Impact of Cancer, and Finding Meaning. The quality of the presented findings is appraised, and implications of the findings and considerations for future research and clinical practice are discussed.

Part Two is an empirical paper, which explores the perceptions of healthcare professionals towards cancer screening and BRCA gene testing for females with a learning disability. Six healthcare professionals were interviewed. Thematic analysis was undertaken to analyse the data. Four themes emerged from the data: Unpreparedness, Role of Others, Communication, and Professional Discomfort. Both the research and clinical implications are discussed.

Part Three is comprised of the corresponding appendices to the systematic literature review and empirical paper. Also included in this is the first author's reflective statement and epistemological statement, which consider the experience of conducting the research and the researcher's values and philosophical position.

Overall word count: 14,104 (excluding tables, figures, appendices and references)

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Part One: Systematic Literature Review

Factors affecting the psychosocial experience of cancer for people with Learning Disabilities

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Please see Appendix A for guidelines for authors.

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Abstract

Background: There are many psychosocial factors associated with health conditions and this is especially true with a cancer diagnosis. People with learning disabilities diagnosed with cancer may face additional challenges during this time compared to the general population. The aim of the current review is to examine the psychosocial factors affecting the cancer experience for people with learning disabilities.

Method: A systematic review of the literature was conducted between March 2019 and January 2020. Papers included in the review were appraised for quality using the National Institute for Health and Care Excellence qualitative checklist (NICE, 2012). The review utilised thematic synthesis.

Findings: Four themes were derived from the data: Communication, Feeling Dismissed, Physical Impact of Cancer, and Finding Meaning. The findings explore the factors affecting the psychosocial experience of cancer for people with learning disabilities.

Conclusions: There appear to be defined factors that affect the psychosocial experience of cancer for people; both positive and negative. The recognition of the psychosocial impact of cancer in people with learning disabilities is important, and the provision of staff education and high-quality support tailored to this patient group are likely to be beneficial.

Key Words: *Learning Disability, Cancer, Experience, Psychosocial*

Introduction

In the United Kingdom, there are approximately 1000 people diagnosed with cancer each day, with prevalence rates and deaths from the disease estimated to rise exponentially by 2030 (Cancer Research UK, 2020; World Health Organisation, 2017). Research has demonstrated that the impact of cancer transcends beyond the more obvious physical aspect of the disease, and extends to the individual's emotional, psychological, and social functioning (Bultz & Carlson, 2006; McFarland & Holland, 2016). People diagnosed with cancer may experience depression, anxiety, difficulties adjusting to their diagnosis, body image issues, loss of valued roles, and perceive themselves as a burden to others (Brintzenhofe-Szoc, Levin, Li, Kissane, & Zabora, 2009; Grassi, Biancosino, Marmai, Rossi, & Sabato, 2007; McPherson, Wilson & Murray, 2007). High quality cancer care is characterised by a holistic attendance to patient needs, and psychosocial care is inherently a significant factor within this (Travado et al., 2017). Existing literature has highlighted the importance of evidenced based psychosocial interventions in reducing cancer related psychological distress, improving quality of life, facilitating self-management, and even potentially influencing survival rates (Galway et al., 2012; Giese-Davis, 2011; Jacobsen, 2009; Spiegel, 2012). Additionally, the NHS Long Term Plan (2019) has prioritised the access to comprehensive care for cancer patients as one of its objectives, with a specific focus on personalised care and information on wider health and wellbeing.

The NHS Long Term Plan (2019) has also committed to improving standards of care for people with learning disabilities in every NHS service. People with learning disabilities experience disparities in healthcare access and provision and suffer higher premature mortality rates than the general population (Hatton & Emerson, 2015). Furthermore, a published literature review of the psychosocial experiences of people with learning disabilities and chronic illness identified that there were substantial gaps in the research

conducted in this area (Flynn, Hulbert-Williams, Hulbert-Williams & Bramwell, 2015). Additionally, the review highlighted evidence of a lack of consideration of the psychosocial needs of people with learning disabilities and chronic illness, resulting in these needs being unmet.

Whilst the average life expectancy for people with learning disabilities is lower compared to the general population, improvements in medical and social care have resulted in people with learning disabilities enjoying increased longevity (Dolan et al., 2019; Emerson & Hatton, 2011; Haveman et al., 2010; Tuffrey-Wijne, 2003). As a result of this, the incidence of age-related illness, including cancer, is rising amongst people with learning disabilities (Sullivan, Hussain, Threlfall, & Bittles, 2004). Rates of cancer in people with learning disabilities are proportionately lower than in the general population, but true figures may be difficult to establish due to missed diagnoses in this population (Baxter & Bradley, 2008).

There is a breadth of literature focusing on aspects of the psychosocial impact of cancer in the general population, however the inclusion of people with learning disabilities within these studies is scant. People with learning disabilities experience difficulties in cognitive abilities such as reasoning, learning, judgement making, planning, and abstract thinking, and in adaptive functioning which includes social functioning, communication, and independent living (Diagnostic and Statistical Manual of Mental Disorders, 5th Edition, American Psychological Association, 2013). These difficulties may present additional challenges to the individual at different stages of the cancer experience compared to the general population. It is therefore important for research to explore the experiences of people with learning disabilities and cancer further, to inform service provision and development for this patient group and ensure that their psychosocial needs are being met.

An existing published narrative literature review has examined the cancer treatment issues for people with learning disabilities (Witham & Haigh, 2018). The review identified that effective communication was important, but challenging to achieve in practice, and this had consequences for decision making. The review also identified that methods of cancer treatment, such as radiotherapy, can be stressful for people with learning disabilities. The review specifically explores the treatment phase of cancer, however the wider literature on psychosocial factors during the cancer experience suggest these are present throughout different stages of the cancer experience, not exclusively during treatment (Grassi Spiegel, & Riba, 2017). The cancer experience has been conceptualised in the literature as “not a single event in time; it is a series of experiences across the continuum of the disease. This continuum extends from the diagnosis to remission and, for many patients, to recurrence and terminal phase” (Zabora et al., 1997, p.74).

The aim of the current review is to synthesise qualitative literature pertaining to the psychosocial experience of cancer in people with learning disabilities. Qualitative literature is preferred in this review to enable the capture of individual experience and personal meaning (Willig, 2013). The principal focus of this review intends to answer the following question: ‘What are the psychosocial factors affecting the cancer experience for people with learning disabilities?’

Identifying commonalities in individuals’ experiences during their cancer journey will be of benefit in facilitating understanding of this patient group’s needs, which is important in ensuring that they receive equitable care. Acknowledging both the individual and systemic factors that influence the experience of cancer in people with learning disabilities may also identify areas of further research interest.

Method

Search Strategy

A systematic search of the literature using electronic databases was undertaken between March 2019 and January 2020. The electronic databases selected for the review were Medline, CINAHL, APA PsycInfo, APA PsycArticles, and Education Research Complete. These databases were selected due to their relevance to the subject area of the review. A grey literature search did not yield any papers for this review. Quantitative papers were excluded as they would not be as conducive to providing accounts of individual experience as qualitative papers. The following search terms were used:

(learning disabilities or intellectual disabilities or mental retardation or learning difficulties or special needs)

AND

(cancer or neoplasms or oncology or tumour or malignancy or carcinoma)

AND

(health care professionals or doctor or nurse or general practitioner or health worker)

AND

(psych* or social)

The search terms used for learning disabilities encompassed both contemporary and historical definitions utilised in existing literature. The inclusion of search terms regarding healthcare professionals was to yield papers with content more pertinent to the context of cancer and the research question.

The following limiters were applied to the search strategy: English language, due to lack of translation resources; and literature published between the years 1990 to 2020. The search terms produced 154 results, which became 123 following the removal of duplicates. Two further papers were removed due to not being in the English language. The titles and abstracts of 121 papers were reviewed for relevance, resulting in 112 papers being excluded. Nine papers were read in full, and following the application of the inclusion and exclusion criteria, the final number of identified suitable articles for this review from the search was seven. Table 1 shows the inclusion criteria and exclusion criteria, with the corresponding rationale for each criterion. The papers which were read in full but not included in the review due to not meeting the inclusion criteria or meeting the exclusion criteria can be viewed in Appendix B. It is accepted that electronic database searches may not return all relevant literature and further manual searches may be necessary (Horsley, Dingwall, & Sampson, 2011). Hand searches yielded one further paper that was suitable for inclusion in the review, thus the final total of papers reviewed was eight. A PRISMA diagram outlining this process is shown in Figure 1.

Table 1. Inclusion and exclusion criteria applied to the review.

Inclusion Criteria	Rationale
Qualitative data	To enable capture of richer data of personal experiences
Including the perspective of an individual with learning disabilities and cancer	The interest of the research is on the experience of cancer for people with learning disabilities
English language	Translation resources unavailable

Exclusion Criteria	Rationale
Quantative data	This would not be conducive to providing accounts of individual experience
Research which did not contain direct quotations or themes.	This would restrict the opportunity for thematic synthesis
Literature focused on cancer prevention or screening issues	The experience of having cancer (from diagnosis, treatment, remission, recurrence to terminal phase) is the interest of the current research
Literature reviews	The current research required original data

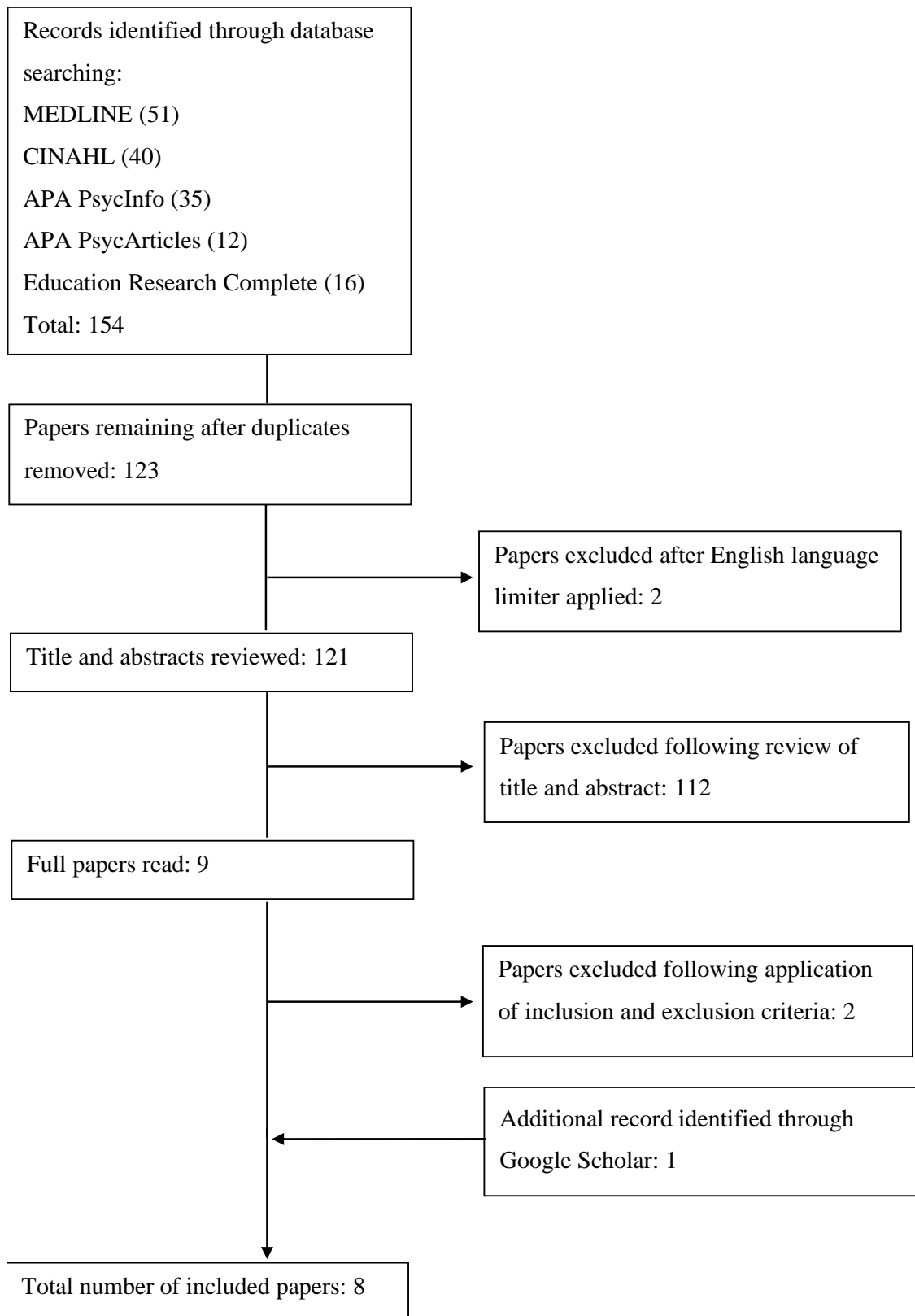


Figure 1. Preferred Reporting Items for Systematic Reviews (PRISMA) flow diagram outlining the search process and outcome (Moher et al., 2009).

Quality Assessment

A quality appraisal of the studies was conducted using The UK National Institute for Health and Care Excellence Checklist for Qualitative Studies (NICE, 2016), as shown in Appendix C. Quality assessment of primary qualitative research has been an area of contention, with much discussion around the appropriateness and usefulness of doing so, and questions around the available tools' reliability and validity (Carroll & Booth, 2015). However, for this review, it was considered important for the overall review's rigour to be aware of the individual papers' relative strengths and limitations. The first author conducted the quality assessment and discussed each rating with the research supervisor. Studies assessed as being of poor quality are excluded by some systematic reviews (Higgins & Green, 2011). However, given the scarcity of research in this area all papers in this review meeting the inclusion criteria were included irrespective of their quality appraisal. Understanding the quality of the papers included in this review remains important and is considered when reporting the rigour of this review and subsequent findings. The quality assessment and rating of each paper included in this review can be seen in Appendix D.

Data Extraction and Data Analysis

The first author developed a tool to extract the data from the papers included in the review (Appendix E). The data extraction tool was developed for extracting data pertaining to the research aims, research methodology, participants, and key findings. Data was synthesised using thematic synthesis (Thomas & Harden, 2008). Thematic synthesis was deemed an appropriate method in identifying themes in the factors affecting the psychosocial experience of cancer across the primary research studies, in order to then create analytical themes guided by an inductive approach (Noblit & Hare, 1988).

The analysis process was comprised of three stages; ‘line-by-line’ coding of participant quotes and researcher themes presented in the original articles’ results section; descriptive themes were then generated across the findings from each study; analytical themes then emerged from the descriptive themes to address the question underpinning the review. Themes were discussed and reviewed with the first author’s research supervisor. Thematic synthesis was preferred over other methods of synthesis for this review and research question as it is specifically developed for reviews that focus on personal experience and perception (Thomas & Harden, 2008).

Results

Characteristics of Included Studies

All eight studies were conducted in the United Kingdom and were qualitative in design. All papers sought to provide an understanding of the experience of people with learning disabilities with cancer. One paper aimed to explore the cancer-related needs of people with a learning disability and conducted interviews with patients which were supplemented by healthcare professional and carer contributions (Flynn, Hulbert-Williams, Hulbert-Williams, & Bramwell, 2016), while another explored the experiences of those who were dying of cancer in an observational study (Tuffrey-Wijne, Bernal, Hubert, Butler & Hollins, 2010). One study focused on disclosure of a cancer diagnosis and people with learning disabilities’ understanding of this, again via observational techniques (Tuffrey-Wijne, Bernal & Hollins, 2010). Tuffrey-Wijne, Bernal, Butler and Hollins (2009) presented the observational findings of an ethnographic study for people with learning disabilities and cancer. Tuffrey-Wijne, Bernal, Jones, Butler and Hollins (2006) utilised observations and semi-structured interviews to explore the cancer information needs for people with learning disabilities who were affected by cancer.

Three of studies included in the review were single case studies of people with learning disabilities who had cancer (Martean, Stedmon & Moss, 2013; Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006). Martean et al (2013) utilised narrative analysis, while the other case studies presented no analysis.

There were 25 participants within the eight studies, however five papers were reporting on the same sample of 13 participants from one core ethnographic study (The Veronica Project) (Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009; Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006).

Across four papers, difficulties in recruitment were reported. Flynn et al (2016) reported that two potential participants were unable to continue taking part due to their illness progressing. The need for 'proxy assent', whereby the individual participant could not provide informed consent for themselves and therefore required a relative or staff member to consent for them, was reported in two papers (Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009). Additionally, across those papers, the authors discuss ongoing 'process consent' to ensure the continued engagement from the participants unable give informed consent themselves, as the data collection occurred prior to the Mental Capacity Act (2005). Tuffrey-Wijne et al (2006) further discuss sampling issues; information on their study was initially presented to support staff prior to people with learning disabilities, and only nine suitable participants were identified in fourteen months. The authors elaborate that recruitment was additionally impeded by eligible participants not being informed of their cancer diagnosis and suggest that this may be due to family and staff attempts to protect them from possible emotional distress. Hence, this precluded their inclusion in the research and as a result only five participants were recruited.

Table 2. Papers included in the review.

Author & Country	Title	Methodological approach	Participants	Summary of Findings	Quality Rating
Cresswell & Tuffrey-Wijne (2008) United Kingdom	The come back kid: I had cancer but, I got through it	Qualitative single case study. No analysis undertaken	1 female (living in an Adult Placement with a couple and their family) with learning disabilities diagnosed with non-Hodgkin's lymphoma. Treatment included chemotherapy and radiotherapy.	The participant's GP did not diagnose cancer despite the participant presenting multiple times with symptoms. Participant collapsed and was taken to hospital, but was not informed by the staff what was happening, resulting in feelings of fear. A second hospital kept her informed and included her throughout in decision making. Participant feels stronger after having cancer. Participant recommends that staff recognise the importance of sharing information with patients, and do this in a way that is accessible to the patient. The participant also references that they would have benefited from having a professional to talk to about their feelings and the impact of the cancer experience upon them.	–
Flynn, Hubert-Williams, Hubert-	“You don't know what's wrong with you”: an exploration of cancer related	Qualitative study adopting semi-structured interviews and grounded theory for analysis	6 participants (all with a mild learning disability): Male aged 53 living with his family, diagnosed	People with learning disabilities can be overlooked during their cancer care and feel excluded. Healthcare professionals often relied upon	++

Williams & Bramwell (2016) United Kingdom	experiences in people with an intellectual disability	<p>with plasmacytoma and bowel cancer. Treatment involved radiotherapy and surgery; Male aged 35 living with parents, diagnosed with testicular and stomach cancer. Treatment included surgery and chemotherapy; Male aged 34 living alone, diagnosed with testicular and stomach cancer. Treatment included surgery and chemotherapy; Female aged 74 living in a residential home, diagnosed with breast cancer. Treatment included a partial mastectomy; Female aged 64 living alone (supported living), diagnosed with breast cancer. Treatment included lumpectomy, preventative double</p>	<p>family members or care staff to facilitate communication, but healthcare professionals who included the patient mediated their distress associated with appointments. Cancer patients with learning disabilities were reluctant to share their distress with caregivers, this led to them appearing cut-off from their experience. Caregivers also appeared to withhold information from the person with learning disabilities to protect them from psychological distress, however this was viewed negatively by the person with learning disability. Providing information to the patient facilitated feelings of empowerment.</p>
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			<p>mastectomy, and chemotherapy; Female aged 61 living in a community group home, diagnosed with ovarian and lung cancer. Treatment was palliative chemotherapy.</p> <p>12 further participants from their supportive network.</p>		
Martean, Dallos, Stedmon & Moss (2013) United Kingdom	Jo's story: the journey of one woman's experience of having cancer and a 'learning disability'	Single case study adopting narrative life story approach, and thematic field analysis	1 female participant, aged 63, with a mild learning disability and breast cancer. Treatment included chemotherapy.	Themes of 'Identity', 'Positioning in Society', and 'Being Positive'. Of particular importance in the latter theme, in which the idea that people with learning disabilities are conditioned to conceal feelings of distress due to others not being able to tolerate them being anything other than happy. Participant reports feeling more confident and empowered following her cancer diagnosis and experience.	++
Tuffrey-Wijne, Bernal, Jones,	People with intellectual disabilities and their	Ethnographic study adopting semi-structured interviews and analysed	5 participants. with a learning disability affected by cancer. All lived semi-independently	The key themes identified were 'Nobody told me'; the desperate need for more information; 'That reminds'; the desire to tell their own	+

Butler & Hollins (2006) United Kingdom	need for cancer information	using grounded theory principles	in their own flats in the community, with paid support staff visiting when needed.	story and 'I don't know much about cancer'. The participants all wanted more information, and with support they were able to comprehend information about cancer. This study highlights the need for accessible resources about cancer and palliative care.	
Tuffrey-Wijne and Davies (2006) United Kingdom	This is my story: I've got cancer: The Veronica Project: an ethnographic study of the experience of people with learning disabilities who have cancer	Single case study adopting thematic field analysis	1 male with mild learning disabilities, diagnosed with penile cancer. Treatment included surgery and chemotherapy, and palliative care,	Participant had an early life experience of doctors disregarding health concerns, which was mirrored in his cancer diagnosis (penile cancer misdiagnosed as a urine infection). Avoidance of seeking medical help or sharing symptoms with his family until symptoms became so severe he was ambulated to hospital. Participant discusses the difficulties associated with telling his family about his cancer diagnosis, and not wanting to upset them. The idea of concealing his true feelings to protect his family are discussed further. He also reflects on the meaning of his illness and the importance he places on his legacy.	-
Tuffrey-Wijne,	People with learning disabilities who have	Ethnographic study adopting semi-structured	13 people with a learning disability and a cancer	The paper highlights the importance of understanding individuals within	++

Bernal, Hubert, Butler & Hollins (2009) United Kingdom	cancer: an ethnographic study	interviews and analysed using grounded theory principles	diagnosis (age range 36-66 years, mean age 53 years).	the context of their lives and past experiences, especially with regards to how this may influence their coping. It also discusses the failure of most doctors in the study to provide information to the patient with a learning disability (N.B. this was before legislation such as the Mental Capacity Act [2005] had been passed into English Law). The lack of treatment offered to some participants in the study warranted further action due to no rationale offered for these decisions, and allegations of discriminatory practice. Participants experienced anxiety and fear which was not acknowledged by healthcare professionals. Doctors appeared reluctant to share the patient's prognosis with them.	
Tuffrey-Wijne, Bernal & Hollins (2010) United Kingdom	Disclosure and understanding of cancer diagnosis and prognosis for people with intellectual disabilities: Findings	Ethnographic study adopting semi-structured interviews and analysed using grounded theory principles	13 participants: Male, 66 with severe learning disability living in a residential home and diagnosed with lung cancer and secondary thyroid cancer. No	11 of the participants had been told about their cancer diagnosis. The themes arising from the research including 'Truth telling' and 'Understanding' were explored in depth, in addition to further exploration of the ideas of 'Is	++

from an
ethnographic study

treatment possible and patient died within nine weeks of diagnosis; Female aged 36 with mild/moderate learning disabilities and living with foster carer, diagnosed with lymphoma. Treatment included radiotherapy and chemotherapy; Male, 44 with a mild learning disability and living in his own flat diagnosed with penile cancer. Treatment included surgery and chemotherapy; Female aged 56 with mild/moderate learning disabilities living with family, diagnosed with breast cancer. Treatment included a surgery and a radiotherapy; Male aged 44 with severe and profound learning disabilities and Down

ignorance bliss' and non-disclosure being a barrier to informed decision making. The paper also offers hypotheses relating to people with learning disabilities' ability to understand a cancer diagnosis being mediated not only by their cognitive ability but their life experiences; their understanding is also further affected by how much they are told and helped to understand.

Syndrome living in a residential care home diagnosed with testicular cancer. Treatment included radiotherapy and chemotherapy; Male aged 47 with profound learning disabilities living in a residential care home diagnosed with cancer. No treatment possible; Male aged 64 with a mild learning disability living on his own flat diagnosed with lung cancer. Treatment included radiotherapy; Female aged 55 with mild/moderate learning disabilities living in a residential care home and then a hospice. Diagnosed with stomach cancer with no treatment possible; Female aged 61 with severe/profound learning disabilities and

Down Syndrome living in a residential care home, diagnosed with breast cancer. Treatment included mastectomy surgery; Female aged 65 with severe/profound learning disabilities living in a residential care home, diagnosed with primary breast cancer and then secondary lung and bone cancer. Treatment included surgery and palliative care; Male aged 47 with mild learning disabilities living independently diagnosed with lung cancer. Treatment included radiotherapy and chemotherapy; Male aged 56 with severe/profound learning difficulties and Down Syndrome living in a residential care home,

			<p>diagnosed with bladder cancer. No treatment possible; Female aged 45 with mild/moderate learning disabilities and autism living alone, diagnosed with primary bowel cancer and secondary cancer in the liver, lung, and spine. Treatment included surgery, radiotherapy and chemotherapy.</p>		
<p>Tuffrey-Wijne, Bernal, Hubert, Butler & Hollins (2010) United Kingdom</p>	<p>Exploring the lived experiences of people with learning disabilities who are dying of cancer</p>	<p>Ethnographic study adopting semi-structured interviews and analysed using grounded theory principles</p>	<p>13 people with mild to severe learning disabilities and a cancer diagnosis (age range 36-66 years, mean age 53 years). Seven participants had adequate verbal communication skills and had some level of independence; six needed continuous support and had severely limited communication skills. Seven participants lived</p>	<p>Main identified themes were ‘Dependent lives’; ‘Deprived lives’; Truth telling and understanding’; ‘The importance of families’; ‘Inexperienced carers, unprepared services’; and ‘Resilience’. This study highlights the importance of understanding the person within the context of their life during their cancer experience. The role and impact and healthcare professionals are also discussed as being important factors in the cancer experience.</p>	<p>+</p>

in staffed residential home, four in their own flat, one in their parental home, and one living with foster parents.

Methodological Quality

Overall, the studies were of varying quality (Appendix D). A qualitative approach was appropriate in all studies.

The papers deemed to be of the highest quality included clear aims, made good reference to existing literature and theory, utilised appropriate data collection to address the research questions, demonstrated details of sound methods of analyses, included detail of external reflective supervision, and clearly identified their roles in relation to the participants in their data (Flynn et al., 2016; Martean et al., 2013; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009.)

Tuffrey-Wijne and Davies (2006) and Cresswell and Tuffrey-Wijne (2008) were both presented as descriptive case studies, and while these papers provide rich data, there were no identified aims or analysis undertaken. There is however an introduction referencing relevant literature in Tuffrey-Wijne and Davies (2006), which is useful in providing theoretical context to the case study.

Three papers present the findings of an ethnographic study (Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009). Different levels of learning disability, age, gender, and social and living circumstances are represented within the studies. However, there is potential bias inherent to the observational techniques utilised by the researchers and this should be considered when drawing conclusions from the papers; for example, the lead researcher across the studies reports a possible effect of her professional background in nursing on her perception when observing participants.

Five papers were reporting on different aspects of the same sample of 13 participants from one core ethnographic study (The Veronica Project) (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009; Tuffrey-

Wijne & Davies, 2006), however there were variations within the methodological quality of each paper.

All papers had a clear statement regarding ethical approvals and considerations.

Synthesis of Findings

Four main themes were derived from the data. These were: Communication; Feeling Dismissed; Physical Challenges of Cancer; and Finding Meaning. Each theme had 2 subthemes (please see Table 3).

Table 3. Superordinate and subthemes derived from the data.

Superordinate Theme	Subtheme
Communication	Feeling informed
	Concealing true feelings
Feeling Dismissed	Delays in diagnosis
	Excluded from own care
Physical Challenges of Cancer	Changes to the body
	Changes to environment
Finding Meaning	Existential purpose
	Personal growth

Communication

Feeling informed

Most papers referenced the importance of feeling informed and understanding what was happening (Creswell & Tuffrey-Wijne, 2008; Flynn et al., 2016; Martean et al, 2013; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2006). This was perceived to be aided by healthcare professionals providing information in an accessible way (Cresswell & Tuffrey-Wijne, 2008) or by making use of published resources about cancer designed for people with learning disabilities (Tuffrey-Wijne et al., 2006). Some patients were able to augment their understanding of their illness by asking medical staff questions (Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010). Information being provided by healthcare professionals to the patient facilitated feelings of greater understanding and empowerment (Flynn et al., 2016), and some patients appeared satisfied with doctors delivering frank information to them (Tuffrey-Wijne et al., 2010). Having sufficient knowledge of their illness enabled the person with cancer to share their diagnosis with friends and family on their own terms, which was considered important to them (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006).

Lack of transparency from medical professionals increased anxiety around feeling uninformed (Flynn et al., 2016; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2006). The use of euphemistic language as opposed to using concrete and easy to understand terminology further exacerbated distress and led to rumination.

'My doctor says that I should go and have a holiday whilst I still can. What is that supposed to mean? Whilst I still can?' (Tuffrey-Wijne et al., 2010, p.17).

The importance of legislative frameworks (such as the Mental Capacity Act, 2005) in ensuring that information is provided to the patient in an accessible way is discussed, and in one study prior to this becoming a legal requirement most doctors failed to provide any information to the patient with a learning disability (Tuffrey-Wijne et al., 2009).

Concealing true feelings

A number of participants across the studies did not want others to know that they were distressed by their cancer, and hence they would avoid disclosing how they truly felt (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010). This pressure to conceal their true feelings was perceived to be an added challenge to their experience of cancer.

'It can be hard. Sometimes I sit in the hospital bed feeling miserable, and I think: 'Right, in the next 10 minutes they'll all turn up to visit me' so I take a deep breath and put a smile on... I've done that plenty of times, make out that I'm alright when inside I'm not, I'm just fed up' (Tuffrey-Wijne & Davies, 2006, p.35)

It appeared that explicitly discussing the experience of emotional distress was accompanied by regret and dismissal of their feelings.

'I'm sorry I'm talking about sad things. You don't want to hear about those do you... I shouldn't have said anything. I'm sorry' (Martean et al, 2013, p.42)

Expressing that one was in physical pain was perceived to be a more acceptable method of eliciting support from others for their cancer related psychological distress.

'I worry or I panic. Sometimes I sit here and there is something on TV that makes me panic, like when they talk about cancer. Then I ring people up and they ask me 'What's wrong?', so I tell them that I'm having pain. I'm not in pain really, I'm just lonely and worried. But I can't keep ringing people up can I?' (Tuffrey-Wijne et al., 2010, p.16)

This reluctance to share their psychological distress may be reflective of people with learning disabilities generally being discouraged by others to share negative emotions and instead present themselves with a more socially acceptable positivity. This has previously

been conceptualised as the ‘handicapped smile’ (Sinason, 1992). People with learning disabilities may not want to jeopardise the care and support that they receive from others, and hence have been conditioned to conceal their feelings of distress and conform to societal expectations of them by means of a ‘forced jollity’ (Tuffrey-Wijne et al., 2010, p.17). The internalisation of this message was particularly evident in a patient account from Martean et al.’s (2013) study:

‘I was told I was very positive and good about the news. The doctors liked me because I didn't make a fuss. John told me that sometimes the doctors think that people like us cause trouble in the hospital’ (p. 287),

Attempts to mediate the feeling of being a burden on others were made by not seeking support, despite this being an evident necessity (Flynn et al., 2016; Martean et al., 2013; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne & Davies, 2006). This effort to conceal feelings of distress appeared to be reciprocated by carers and relatives of people with learning disabilities and cancer. Studies reported that paid caregivers and relatives appeared unable to tolerate their own feelings of discomfort which consequently further inhibited the ability of the person with a learning disability to share any feelings of distress (Flynn et al., 2016; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009). It appeared that this bi-directional process of avoiding difficult conversations was a significant barrier to mutual understanding and communication. The provision of professional support pertaining to the psychological distress for the person with cancer was identified as a desired resource that was unfortunately absent (Cresswell & Tuffrey-Wijne, 2008).

Feeling Dismissed

Delays in diagnosis

Difficulties in receiving a cancer diagnosis in a timely manner were evident across the studies. This was particularly apparent for individuals who presented to their GPs with symptoms and had their concerns wholly dismissed or misattributed to less serious conditions (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2009). Despite repeated attendances to the GP and on one occasion with support from a social worker, one study demonstrates multiple instances of the individual's concerns not being respected (Cresswell and Tuffrey-Wijne, 2008). Another study details one man living independently presenting to his GP and erroneously being given antibiotics for a urine infection; however, his reluctance to inform his family, and his fear of symptoms, medical staff and hospitals prevented him from seeking further help (Tuffrey-Wijne & Davies, 2006). In both cases, diagnosis only occurred following the individuals deteriorating significantly and requiring an emergency hospital admission.

People with learning disabilities were often dependent on others, such as relatives or carers, in noticing or escalating concerns around symptom development. Instances of the person with learning disabilities being accused of attention-seeking, or care staff not seeking medical advice despite obvious deterioration in their physical health also contributed to delayed diagnoses (Tuffrey-Wijne et al., 2009). Advanced disease progression and metastasis were consequences of delays in diagnosis and led to poorer outcomes, such as extensive surgery, amputations, and death (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2009).

Excluded from own care

People with learning disabilities were often excluded from being an active participant in their own care. This extended to not being acknowledged by medical staff (Tuffrey-Wijne et al., 2010), staff not using accessible terminology (Cresswell & Tuffrey-Wijne, 2008) and

medical professionals speaking another language that was not familiar to the patient whilst discussing their care (Flynn et al., 2016).

'You don't know what's wrong with you... I can't understand what they... the words that they're saying' (Flynn et al., 2016 p.1201).

Medical staff appeared reluctant to share the patient's prognosis with them, and decisions were often made without the involvement of the patient, but rather with carers (Tuffrey-Wijne et al., 2009). Feeling excluded led to an increase in psychological distress and anxiety around what was happening, and this was often not recognised by healthcare professionals (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne et al., 2010). Moreover, paid caregivers' or relatives' efforts to protect the patient from knowing about their health were seemingly misplaced and viewed negatively by the individual (Flynn et al., 2016). Attempts to engage the patient meaningfully with their care was regarded positively within studies (Flynn et al., 2016; Cresswell & Tuffrey-Wijne, 2008) but did not appear to be a common occurrence for the participants across the studies.

'Then there were lots of meetings...It was good that they explained all my options to me...And he asked me what I wanted'. (Cresswell & Tuffrey-Wijne, 2008, p.154)

Feeling excluded during their cancer care seemingly mirrored a history of people with learning disabilities being excluded more generally in society, as reflected in one paper:

'You get it a lot when you're like me. Usually they talk to John and don't talk to me. It's because I have a learning disability' (Martean et al., 2013, p.286).

Physical Challenges of Cancer

Changes to the body

The physical effects of cancer and its treatment were identified as having a negative psychosocial impact across the studies, especially if the symptoms and disease had become advanced (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2009). There was a sense of horror towards the extent of participants' untreated physical symptoms:

'[My brother] saw this lump, and this stuff coming out of it...It made him feel sick'. (Tuffrey-Wijne & Davies, 2006, p.8).

'The urology specialists at the hospital were shocked...they had never seen such an advanced case of penile cancer before' (Tuffrey-Wijne et al., 2009, p.505)

Pain, significant weight loss, fatigue, breathlessness and haemoptysis were symptoms documented across the studies (Cresswell & Tuffrey-Wijne, 2008; Flynn et al., 2016; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009).

Changes to the body were often characterised by loss; this included physical loss from surgery, whereby one man had his genitals, bladder and bowel removed; loss of strength and mobility; and chemotherapy related hair loss (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009). Loss of engagement with previously enjoyed recreation was also evident, though some people still chose to attend their day centres despite not being well enough to participate in activities (Tuffrey-Wijne et al., 2010).

The psychological impact of appearance changes or loss of sexual organs were not explored in depth across the studies and may be reflective of the assumptions that these issues are not considered important for people with learning disabilities (Gomez, 2012). Furthermore, three women who had undergone mastectomies were not offered reconstructive

surgery, which may again illustrate this assumption (Tuffrey-Wijne et al., 2009). Physical changes appeared to be a source of fear and frustration, particularly if the person had not been helped to understand their disease progression:

'[She] was never told that she was dying. She became increasingly distressed about her weakening body, fruitlessly trying to exercise her legs' (Tuffrey-Wijne et al., 2009, p.506)

Concerns around the stigma of cancer symptoms and treatment side effects were evident, with one participant expressing distress at having been accused of not practising good hygiene prior to his diagnosis, and being laughed at when vomiting in public following chemotherapy (Tuffrey-Wijne & Davies, 2006). Being advised about the physical side effects of treatment helped mediate the distress associated with them (Cresswell & Tuffrey-Wijne et al., 2008).

Changes to environment

Changes to the individuals' physical environment necessitated by their cancer was demonstrated across the studies. This included having to leave their own homes to receive treatment in hospital or receive palliative care in a hospice, or relocating to relatives' homes for additional support (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009). There were challenges associated with this, such as familiar care staff unable to remain with an individual in hospital due to having other residents to care for. This resulted in the patient being alone in hospital, with hospital staff that struggled to understand them (Tuffrey-Wijne et al., 2009). Additional challenges came from other patients on general hospital wards who could be abusive towards the patient with a learning disability (Cresswell & Tuffrey-Wijne, 2008).

For those that could remain living independently, adaptations were needed to facilitate this, such as relocating to a ground floor property due to no longer being able to climb stairs. Delays in securing suitable housing resulted in placements into older adult residential homes for some participants, which was inappropriate due to them being significantly younger in age (Tuffrey-Wijne & Davies, 2006).

Finding Meaning

Existential purpose

People with learning disabilities and cancer appeared to undergo a process of reframing their cancer experience within the context of a higher purpose (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006). The desire to impart their knowledge to healthcare professionals to improve care for future patients was apparent. This also included wanting to share their stories with others to ensure that people with learning disabilities had a greater awareness of cancer symptomology and to not suffer as they had. Individual suffering was noted to be conceptualised as having a greater meaning and to help the individual come to terms with their disease and prognosis:

'I think I was put on planet earth to help people. So if my story can stop someone from getting cancer...If that helps, then I am ever blessed' (Tuffrey-Wijne & Davies, 2006, p. 10).

'I really hope that other people can learn from all this. I want to help others' (Cresswell & Tuffrey-Wijne, 2008, p.155).

The participants across the studies had a shared commonality of experiencing adversity more broadly in their lives, such as untreated poor physical health in childhood, bereavement, abuse, enforced sterilisation and being subjugated in society (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et

al., 2010; Tuffrey-Wijne et al., 2009;). The sense of being ‘experienced sufferers’ (Tuffrey-Wijne et al., 2010, p.17) in life appeared to evolve into a position of power for some participants who seemingly restructured their life and experience by ascribing positive meaning to it following their cancer diagnosis (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006). This was however dependent on the level of the individual’s learning disability, and how much support they had been given around understanding their diagnosis by medical staff, or carers and relatives (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006).

Personal growth

The experience of cancer for the individual was identified as a catalyst for personal growth across a number of the studies (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009). Participants across the studies reported greater acceptance, developing confidence and feeling more motivated as a result of cancer, despite their learning disability; this was particularly evident in the case studies (Cresswell & Tuffrey-Wijne, 2008; Martean et al., 2013):

‘Having cancer has definitely made me stronger. Before, sometimes I couldn't be bothered to do something I'd think “oh I'll do it tomorrow or the next day” ...I've been given a second chance’ (Cresswell & Tuffrey-Wijne, 2008, p. 156).

Discussion

Overview

The present review aimed to identify the psychosocial factors affecting the cancer experience for people with learning disabilities. All papers adopted a qualitative approach,

and included perspectives of the individuals with cancer, and others involved in their care. There were disparities between the quality ratings of the papers.

The themes apparent in the review appear relatively consistent with factors affecting the cancer experience identified in the broader psycho-oncology literature. Feeling informed about one's illness emerged as an important factor for people with learning disabilities and is similarly recognised as a key factor in the cancer experience for the general population (Martinez, Schwartz, Freres, Frazee & Hornik, 2009). Within this review, participants were described as having a dependence on others to provide information about their illness and aspects of their care, frequently as a result of not having this information provided directly to them. Cancer patients in the general population may be more able than those with learning disabilities to independently seek resources to meet their information needs, such as through asking questions to professionals directly, or accessing online searches and other existing literature. (Eysenbach, 2003). Only one paper included the use of adapted materials in explaining the implications of a cancer diagnosis to people with learning disabilities (Tuffrey-Wijne, Bernal, Jones, Butler & Hollins, 2006). Feeling excluded as an active participant of their care was also apparent; this is not in accordance with existing literature emphasising the importance of effective communication in cancer care (Fallowfield & Jenkins, 1999). Informed and included patients feel more empowered which can mediate some of the cancer related distress (Mesters, van den Borne, De Boer & Pruyn, 2001), and within the papers reviewed it was evident that people with learning disabilities and cancer could be helped to understand their illness with necessary support, and this was received positively and helped alleviate distress.

People with learning disabilities may be more accustomed to receiving care and support from others (Tuffrey-Wijne et al., 2010). Despite this, it was apparent that there were concerns of being perceived as a burden by others during their cancer experience. The sense

of feeling like a burden is discussed within existing literature for the general population during their cancer experience, which may increase distress and inhibit help-seeking (McPherson et al., 2007). However, there may be a more fundamental avoidance of expressing distress in people with learning disabilities; the ‘handicapped smile’ has been conceptualised as a conditioned response in people with learning disabilities being discouraged from sharing negative feelings due to the discomfort this may cause others (Sinason, 1992), and this reluctance to share their distress was evident from the review. Throughout the papers, the level of learning disability is not always stipulated, and hence ability level may dictate the participants’ level of care and choice around help-seeking.

Delays in obtaining a cancer diagnosis can result in poorer outcomes and increase the possibility of more invasive interventions further along the disease trajectory (Neal et al., 2015). Whilst delays in diagnosis are not an exclusive issue for people with learning disabilities, they are at greater risk of this happening due to poorer health literacy (Elliot, Hatton & Emerson, 2003), and the significant role of others in escalating their health concerns and facilitating presentation to healthcare services (Alborz, McNally & Glendinning, 2005). Within this review, delays in diagnosis appeared to stem from negative perceptions from key stakeholders in the person’s care, and ‘diagnostic overshadowing’, whereby genuine healthcare complaints were dismissed or attributed to an individual’s learning disability (Mason & Scior, 2004). Feeling dismissed thus had both negative physical and psychological consequences, across this review.

The physical changes that may accompany a cancer diagnosis have been demonstrated to affect the psychosocial experience of cancer (Grassi, Biancosino, Marmai, Rossi & Sabato, 2007). Changes to physical appearance as a result of the disease and treatments can have negative psychological consequences (Fingeret, Teo & Epnet, 2014). Furthermore, surgical removal of body parts associated with sexual functioning and sexuality

may cause further psychological challenges, in terms of how one views themselves as an individual, within the context of relationships, and even within society (Burbie & Polinsky, 1992; Fobair et al., 2006). However, across the papers included in the review, there appeared to be little consideration of this for people with learning disabilities. This may demonstrate the lack of consideration that is afforded to people with learning disabilities' values around body image or relationships, and the assumptions that others may hold around this issue (Aylott, 1999). People without learning disabilities and cancer are also more likely to be able to remain in their own home. It was identified in this review that people with learning disabilities could be inappropriately moved to residential homes for older adults, which again may be reflective of the lack of consideration of their needs from others, and assumptions that this would be acceptable.

In addition to cancer's negative psychosocial sequelae, there may be positive aspects of the experience. Post-traumatic growth and finding meaning in cancer have been explored in the broader literature, and the findings suggest that some cancer patients are able to frame their illness positively (Stanton, Bower & Low, 2006). This idea emerged from the present synthesis in the belief that their existence and experience served a higher purpose, and the personal growth they achieved. This was present for both cancer survivors and those who were aware that they were in the terminal phase of their illness (Cresswell & Tuffrey-Wijne, 2008; Martean et al, 2013; Tuffrey-Wijne & Davies, 2006). This theme was however more apparent from individuals with higher levels of cognitive abilities.

Limitations

The present review yielded eight papers, and whilst this is deemed sufficient for thematic synthesis (Thomas & Harden, 2008), the features of the included papers may impose some limitations on the conclusions that can be drawn from the review. Two papers did not

obtain a favourable quality appraisal due to no identified research aims, no analysis, and no discussion of the content (Creswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006); however, the case study design adopted in these papers allowed for a rich description of their cancer experience. The case study approach also allows for the voice of this group to be heard in a society where they may face challenges of powerlessness (Johnstone, 2018).

Moreover, as five papers were reporting on the same sample of 13 participants from one core ethnographic study (The Veronica Project) (Cresswell & Tuffrey-Wijne, 2008; Tuffrey-Wijne & Davies, 2006; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2010; Tuffrey-Wijne et al., 2009), it is important to consider the potential effect this has on the overall review. Multiple 'salami publication' of papers may result in an over-emphasis of the results from one study, thus limiting the extent to which the overall findings can be viewed with confidence (Kassirer & Angell, 1995). In the context of this review, it may potentially jeopardise how representative these findings are of the psychosocial experience of people with learning disabilities during the cancer experience more broadly. However, these studies did provide a good insight, and were worthy of inclusion. The ethnographic design present in the majority of the included studies lends itself to researcher interpretation, and as such the presented results are likely to be through the lens of the researcher, despite efforts to mediate this (LeCompte, 1987).

All included studies had been conducted in the United Kingdom. This may present some bias in terms of approaching the understanding of learning disabilities from a Eurocentric position, and with regards to how healthcare is structured within the National Health Service.

Only two papers in the review had been conducted and produced since 2010, and so this may not fully encapsulate more contemporaneous experiences or reflect current socio-

political factors that can affect healthcare; for example, one study included in this review had been undertaken prior to the Mental Capacity Act (2005) and others had been published prior to the Equality Act (2010). The recent increase of learning disability liaison nurses, a role in which the gap between healthcare and specialist learning disability care is bridged, has been reported to positively impact people with learning disabilities in hospitals, and therefore may affect the cancer experience for this patient group, again demonstrating the need for more contemporary research (Castles, Bailey, Gates & Sooben, 2014).

Research and Clinical Implications

The limited numbers of papers yielded during the review demonstrates a dearth of research interest in this area, which may be a wider reflection of the themes of dismissal and exclusion highlighted in this review. It also demonstrates that the population of people with learning disabilities and cancer as research participants is one that is hard to access, and may explain why the same sample was used across multiple publications included in this review. People with learning disabilities not being included as active research participants may be as a result of the paternalistic care this group receives, as highlighted in this review's findings. Where people with learning disabilities and cancer are supported to be directly involved with research, their contribution can be meaningful and well-regarded (Martean et al., 2013). Robust future research into their experiences of cancer directly could help establish a greater understanding of their needs and how to best support them. Further, the effectiveness of psychological or psychosocial interventions for people with learning disabilities and cancer is an area that warrants further research attention. One paper specifically identified that the provision of professional psychological support would have been an appreciated resource to help with the experience (Cresswell & Tuffrey-Wijne, 2008). The need for a space to talk about the effect of cancer outside of the individual's immediate support systems was also apparent; the reluctance to share distress appeared to directly relate to the impact this would

have on how others familiar to them would perceive them. This may be mediated by an external provider of support, for example, a Macmillan nurse or a Psychological Medicine service making reasonable adaptations for a patient with learning disabilities. Research has demonstrated that people with learning disabilities are able to engage with therapeutic interventions (Haddock & Jones, 2006; Hodges & Sheppard, 2002). Adapting interventions around death and dying that are available for the general population for cancer patients for people with learning disabilities may be useful for the individual in understanding their illness and encouraging quality of life at end of life. Furthermore, supporting the individual with a learning disability beyond cancer may be beneficial, in terms of delivering interventions designed to allay fears of recurrence, and even promoting post-traumatic growth, as evident in the wider psycho-oncology literature and reflected in the case studies included in this review (Creswell & Tuffrey-Wijne, 2008; Martean et al., 2013; Tuffrey-Wijne & Davies, 2006). Further support for relatives and care staff may also be beneficial in considering the importance of transparency and managing conversations that may be challenging.

The role of healthcare professionals and carers or relatives should also be an area of further research consideration, as it was apparent from the review their influence in the factors affecting the cancer experience for people with learning disabilities. Understanding their perceptions may help identify areas for intervention and thus improve the patient experience. The NHS Long Term Plan (2019) has identified that all NHS staff should be educated on the needs of people with learning disabilities and supported to make reasonable adjustments in services. Further specific education for medical staff pertaining to the psychosocial impact of cancer for people with learning disabilities is likely to improve their approach to working with this patient group.

Conclusions

People with learning disabilities appear to experience similar psychosocial effects of cancer as people without, and the findings of this review appear consistent with the broader psycho-oncology literature. There are however some pronounced differences in the factors affecting this experience for people with learning disabilities; most notably the feelings of exclusion from their own care, and the dominant role of others. Further high quality, contemporary research is necessary to understand the cancer experience of people with learning disabilities with this specific population as active participants. As key stakeholders to this experience, healthcare professionals' perceptions should also be explored further at different stages of the cancer experience.

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Part Two: Empirical Paper

An exploration of healthcare professionals' perceptions towards testing for cancer in females with learning disabilities

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Please see Appendix A for guidelines for authors.

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Abstract

Background: People with learning disabilities can experience inequities in healthcare, and this is especially true for women. Cancer screening can help with early detection, prophylactic intervention, and better prognosis, however females with a learning disability may not have adequate rates of access to these screening programmes. Staff perceptions towards specific patient groups has previously been linked to differences in care and equitable access to services. The aim of the current study was to identify themes in the perceptions of healthcare professionals towards cancer screening and BRCA gene testing for females with a learning disability.

Method: Using a qualitative approach, semi-structured interviews were carried out with six healthcare professionals. Interviews were transcribed and the data were analysed using Thematic Analysis.

Findings: Four themes emerged from the data: Unpreparedness, Communication, Role of Others, and Professional Discomfort.

Conclusions: Healthcare professionals expressed a desire for more teaching on working with patients with a learning disability during their training. They recognised the importance of ensuring the person with a learning disability was helped to understand procedures, and the role of family members, carers, and the wider system. It may be beneficial for healthcare professionals to be offered time to engage in reflective practice for the emotional aspect of their role.

Key Words: *Learning Disability, Cancer screening, BRCA testing, Healthcare Professionals, Perceptions*

Introduction

A learning disability is defined in the Diagnostic and Statistical Manual-V as the presence of difficulties in cognitive abilities such as reasoning, learning, judgement making, planning, and abstract thinking, and in adaptive functioning which includes social functioning, communication, and independent living (DSM-5; American Psychological Association, 2013). There are thought to be approximately 1.5 million individuals with a learning disability living in the United Kingdom, with an estimated 375,000 of this population being a female aged over 18 years (Emerson et al, 2012).

The occurrence of premature deaths in people with learning disabilities as a result of not receiving appropriate healthcare has been demonstrated in the literature (Krahn, Hammond & Turner, 2006). The charity Mencap's 'Death by Indifference' publication (Mencap, 2007) highlighted that within the NHS, people with learning disabilities were not being adequately cared for, and medical and nursing staff seemed unaware of their responsibilities with regards to complying with the Mental Capacity Act (2005). Following this, The Confidential Inquiry into Premature Deaths of People with Learning Disabilities (CIPOLD; Heslop et al, 2013) in England was commissioned to provide evidence about contributory factors to avoidable and premature deaths in this population. Of note, the report detailed the disparity in life expectancy between females with a learning disability and those without; women with learning disabilities died on average 20 years younger. It also highlighted that women with learning disabilities had shorter life spans on average than men with learning disabilities; this is in contrast to the general population, where women have greater life expectancy than men on average (Austad, 2006). One of the contributing factors to these issues was the lack of reasonable adjustments made for patients with learning disabilities, particularly during their attendance at clinic appointments. GP referrals

commonly did not contain information about the needs of the patient or state the person had a learning disability. Again, the results found that healthcare professionals demonstrated a lack of understanding of the Mental Capacity Act (2005); particularly regarding assessing capacity, making best interest decisions and the use of Independent Mental Capacity Advocates in safeguarding the individual with learning disabilities.

The inquiry also identified cancer as the cause of approximately 20% of the deaths recorded for people with learning disabilities. In response to CIPOLD, since 2016 there have been yearly Learning Disabilities Mortality Reviews (LeDeR; Heslop et al, 2016; 2017; 2018; 2019) The most recent LeDeR findings highlighted the lack of support in accessing cancer screening. The NHS cervical screening programme in England is offered to females aged from 25 to 64. Routine screening is offered every three years up to 49 years of age and every five years from 50 to 64 years of age (NHS Cervical Screening Programme, 2015). Breast screening is offered to females aged 50 to their 71st birthday in England (NHS Breast Screening Programme, 2015). Screening and early detection can lead to a better prognosis and reduce the need for more invasive interventions further along the disease trajectory (Parmeshwar, 2018). Breast cancer is the most common cancer in the UK, with incidence rates projected to rise even further by 2035 (Cancer Research UK, 2020). There are approximately 3200 cervical cancer diagnoses each year in the UK, with many strains of cervical cancer caused by the Human Papilloma Virus, which can be contracted through sexual activity (Cancer Research UK, 2015; Crosbie, Einstein, Franceschi & Kitchener, 2013). Inequities faced by females with a learning disability in accessing cervical and breast screening has been further evidenced in existing research (Cobigo et al, 2013). Figures suggest that the incidence of breast cancer is lower in females with learning disabilities than in the general female population; however, it is thought that this data may be skewed by

lower rates of breast screening uptake and missed diagnoses in females with a learning disability (Sullivan et al, 2003). Lower levels of cervical cancer reported in females with a learning disability is also possibly due to the disparities in accessing cervical screening; 31% versus 73% screening uptake in women with learning disabilities and without, respectively (Primary Care Domain, 2019). Assumptions around the sexual activity status of women with learning disabilities may also result in cervical screening not being considered a priority for them to attend (Heslop et al, 2019).

Female specific healthcare for women with learning disabilities has been discussed in the literature; Willis (2015) explored the reasons for attendance and non-attendance to mammograms using a qualitative method to investigate themes. The results suggest that reasons for non-attendance were concerns about pain, lack of awareness, and lack of understanding in the absence of accessible information being presented. Practical reasons such as not being able to read appointment letters and difficulties with travelling to appointments were also highlighted. Reasons for attending were thought to be as a result of previous or current breast illness, or possible differences in the support available from family or carers. Additionally, one participant with a heightened hereditary risk of breast cancer was not offered a prophylactic mammogram, and this was not pursued by their carer despite them knowing the risk. This again reflects the passive position of females with learning disabilities in accessing healthcare if they are not supported and empowered, and the importance of the role that others take in facilitating this. This study provides good insight into the experiences of women with learning disabilities either attending or not attending breast screening appointments from their own perspective. Research into the experiences of women with learning disabilities attending cervical screening has also been explored; Lloyd and Coulson (2014) assessed the difficulties experienced, finding that barriers to accessing cervical

screening for women with learning disabilities included lack of awareness, negative beliefs and attitudes towards having the procedure done from the individuals themselves.

Furthermore, healthcare staff attitudes were also implicated as deterrents for the women attending. This was particularly with respect to the staff not having experience of working with this population and thus not demonstrating the necessary attitudes and skills conducive to making them feel less distressed during the procedure. This study also highlighted the importance of learning disability nurses in facilitating education around the procedure for the patient, and in strengthening joint working with the staff performing the procedures.

Females with a family history of breast or ovarian cancer can access further screening procedures. Mutations in the breast and ovarian cancer susceptibility (BRCA) genes 1 and 2 greatly increase a woman's risk of developing hereditary forms of breast and ovarian cancer at a younger age (King, Marks & Mandell, 2003). Predictive testing for these mutations is possible by analysing a blood or saliva sample from the individual at possible risk of prematurely developing a hereditary cancer. A positive result indicates that the woman has the genetic mutations which increase the risk of developing these forms of cancer. The incidence rates of cancer developed as a result of BRCA gene mutation is thought to be around five percent to ten percent in breast cancer diagnoses, and ten to fifteen percent of ovarian cancer diagnoses (Royal Marsden, 2016). It is estimated that of the population of females carrying the BRCA gene mutations, fifty percent will develop breast cancer and thirty percent will develop ovarian cancer before the age of 70, with the majority developing before age 50 (King et al, 2003). At present, a review of the published research of BRCA testing in females with learning disabilities yields only one result that describes this process with one woman with 'mild mental retardation' (*sic*), in the context of a peer supervision dilemma (Schneider, Keiffer & Patenaude, 2000). With the exception of Schneider et al

(2000), there currently exists no published research that explicitly studies genetic testing for BRCA1 and BRCA2 mutations in females with learning disabilities.

The presence of a learning disability may be implicated in the scarcity of research in this area. Difficulty with decision making is characteristic of a learning disability (DSM-V; American Psychological Association, 2013). This has been reflected in research examining capacity in people with learning disabilities when making a health care related decision (Wong, Clare, Holland, Watson & Gunn, 2000). Consent is an important factor underpinning all aspects of health care and this is especially imperative for intimate healthcare procedures, such as cervical screening and breast screening (Griffith, 2009). Having capacity is an integral part of giving informed consent, and a review of the literature emphasises the importance of the attitudes and behaviours of healthcare professionals in facilitating this process in people with learning disabilities (Goldsmith, Skirton & Webb, 2008). When compared to other groups, adults with a learning disability require the most support to ensure that they are making a decision that is within their capacity to make, or when capacity is not deemed to be present, decisions made by others, usually support staff or healthcare professionals, acting in the individual's best interest are necessary (Mental Capacity Act, 2005).

The Social Model of Disability proposes that the concept of disability is one that has been socially constructed, and that individuals themselves are not inherently disabled but rather society is organised in a way that is detrimental to those with impairments (Oliver, 2013). This can be identified more obviously with structural barriers for people with physical disabilities, for example buildings lacking ramps for wheelchair users, or people with learning disabilities not being given important information in an accessible format. However,

for people with learning disabilities, other factors that can be disabling are less salient. The latter has been described in the literature as ‘psycho-emotional disablism’, which can impact on the individual with a learning disability’s sense of self and well-being (Thomas, 2004). Psycho-emotional disablism can operate at an individual level, for example by a medical professional causing distress by failing to provide information regarding healthcare to an individual with learning disability in an accessible way, resulting in feelings of confusion and frustration (Reeve, 2014). This can also occur at an institutional level, such as high profile cases of systemic abuse within services, and even within the NHS with respect to people with learning disabilities’ mortality rates when compared to the general population (Heslop et al, 2013). Sinason (2002) discusses the psychological impact this can have on people with learning disabilities if they internalise societal messages that can undermine the value of their lives, describing this experience of ‘unwantedness’ and societal rejection as another form of oppression (Sinason, 2002, p.39).

Such societal narratives can affect how healthcare professionals interact with certain patient groups. For example, research into the attitudes of oncology professionals towards older people have demonstrated that negative attitudes towards this patient group exist, and hence can permeate into clinical care, especially with regards to treatment intended to aid life longevity in older people (Kearney. Miller, Paul & Smith, 2000). Previous quantitative research into the attitudes of GPs towards patients with a learning disability identified that negative attitudes were not reflected as explanations for discrepancies in care, and that wider factors related to their perceptions of this specific patient group were contributing to this (Gill, Kroese & Rose, 2002). Perception can influence an individual’s opinion, judgement, understanding and response to people and situations, and the meaning they ascribe to experience (Munhall, 2008).

The aim of this research is to identify themes in the perceptions of healthcare professionals towards cancer screening and BRCA gene testing for females with a learning disability. At present there is a paucity of research in this area, despite there being evidence of disparities in cancer screening uptake for women with learning disabilities. A modest amount of research focusing on the experience of females with learning disabilities undergoing cancer screening procedures exists, highlighting some of the challenges they face and often referring to healthcare staff's role within this. A qualitative approach will be undertaken to obtain staff perceptions of performing cancer screening for this patient group and will then be thematically analysed. The emergent themes could be considered beneficial towards identifying the needs of staff in supporting this patient group. More broadly, this research aims to contribute to the literature on healthcare for people with learning disabilities, and more specifically to contribute to the relatively scarce existing literature on cancer screening in women with learning disabilities. Therefore, the question underpinning this research is 'What are the perceptions of healthcare professionals towards cancer screening and BRCA gene testing for females with a learning disability?'

Method

Design

The study employed a qualitative design using individual semi-structured interviews with healthcare professionals to generate data and address the primary research question. Focus groups had been initially considered, however due to the professional backgrounds required of participants there may have been difficulties in co-ordinating the group for a time convenient for all. Furthermore, differences in potential participants' professions may have resulted in hierarchical issues and may not have fully encapsulated all participant's views (Grønkjær Curtis, de Crespigny & Delmar, 2001). Lastly, individual interviews typically

yield longer speaking times than individual participant contribution in focus groups and therefore may provide more insight into their perceptions (Adams, 2010). Thus, individual interviews were preferred. The interview schedule utilised open ended questions with prompts and can be viewed in Appendix F. Data generated by the interviews were extracted using thematic analysis (Braun & Clarke, 2006). Other methods of data analysis considered for this study included grounded theory (Glaser & Strauss, 1967) and Interpretative Phenomenological Analysis (Smith, Flowers & Larkin, 2009); however, both were rejected due to these methodologies not fitting the research aim. The researcher approached the thematic analysis with an inductive approach, allowing themes to emerge from the data (Patton, 1990) rather than attempting to fit the data within an existing frame or in the researcher's preconceptions (Braun & Clark, 2006).

Participants

Participants were recruited by means of a speculative email included in the global mailing newsletter update to staff in NHS Trusts geographically close to the researcher. The email contained information regarding the inclusion criteria, the time requirements, and the researcher's email address and contact number (Appendix G). Social media advertisement was utilised for further research promotion and participant recruitment (Appendix H). Following their expression of interest in taking part in the study, participants were provided with an information sheet detailing the research aims and what their participation would involve (Appendix I). If meeting the inclusion criteria and satisfied with the information sheet, participants provided their written informed consent and returned this to the researcher (Appendix J).

Participants were deemed to meet the inclusion criteria for participation if they were proficient in speaking English; a healthcare professional registered with a professional

regulatory body; and had experience of providing cancer screening. Due to the COVID-19 pandemic, there were some difficulties in the recruitment of healthcare professionals. Six healthcare professionals from four different NHS Trusts in England were recruited to take part in the research; three were employed by a local NHS Trust and three were recruited from social media and employed by three separate NHS Trusts. Sample sizing in qualitative research is an area of conceptual debate (Vasileiou, Barnett, Thorpe & Young, 2018). It has previously been recommended that qualitative studies obtain a sample size of at least twelve to achieve data saturation (Braun & Clarke, 2013). However, more recent guidance has concluded that the prevalence of themes can be identified within six interviews (Guest, Namey & Chen, 2020). In this study, six participants were deemed sufficient to reach thematic saturation as four clear themes had been identified across the interviews (Glaser & Strauss, 1967).

In order to protect confidentiality and avoid ‘deductive disclosure’, specific participant demographics will not be included, and pseudonyms will be assigned to participant quotes (Kaiser, 2009). Participants were all female and with roles and experience in primary care and oncology services. The professional backgrounds represented in the study were nursing and physician associate. Length of participant experience of working in their qualified roles in the NHS ranged from one year to 28 years (mean=13).

Procedure

Ethical approval for this study was granted by the University Research Ethics Committee, and approval for using NHS staff in the study was obtained from the Health Research Authority (see Appendix K). Face to face interviews have been described in the literature as the “gold standard” for qualitative research (McCoyd & Kerson, 2006, p. 389). As a consequence of the national COVID-19 restrictions, it was not possible to attend sites

nor conduct in person interviews. Therefore, all interviews were conducted via telephone, and audio recorded with participant permission. Telephone interviews have been subject to some criticism with regards to the potential loss of visual cues and non-verbal nuances which may elicit further conversation in interviews (Garbett & McCormack, 2001). However, a review by Novick (2008) concluded that there was little evidence to suggest that telephone interviews compromise the quality of obtained data, and further argue that in person interviewing may create interpersonal stressors that are not present for telephone interviews.

Prior to the interview commencing, participants were advised of the sensitive nature of the research content and that the interview could be paused or terminated; information pertaining to sources of support was given to participants following the interviews (Appendix L). Participants were also informed of the researcher's responsibility towards escalating any evident safeguarding issues. Participants were at liberty to withdraw their data from the study up until the point of data analysis and given a time frame of one month for this.

Data Analysis

Thematic analysis was used to analyse the data generated from the interviews. Thematic analysis is useful in examining the perspectives of different research participants and allowing unanticipated findings to emerge (King, 2004). Thematic analysis was also selected due to the theoretical flexibility it offers (Braun & Clark, 2006), which was considered important in relation to the exploratory nature of the research. An inductive analytical approach was taken, guided by the six-stage analysis procedure outlined by Braun and Clark (2012). The researcher's epistemological stance can be viewed in Appendix O. Table 1 shows the phases and processes conducted. A worked example of the data analysis can be seen in Appendix K. Côté and Turgeon (2005) recommend obtaining participant

feedback on the analyses; however, this was not possible in this study due to time and funding constraints.

Table 1. Phases and Processes of Thematic Analysis (adapted from Braun and Clarke, 2012).

Phase	Process
1. Familiarisation with data	Using the transcribing process to listen to the data, reading and re-reading the transcripts noting initial ideas.
2. Generating Initial Codes	Systematically coding key features of the data, and collating data relevant for each code.
3. Searching for Themes	Collating codes into potential themes and gathering data relevant to the theme.
4. Reviewing Themes	Establishing if themes work in relation to the extracted codes and are relevant to the data set.
5. Defining Themes	Further refining of each code; generating clear definitions and names for the themes.
6. Producing the Report	Writing the report, selecting appropriate quotes for each of the relevant themes and integrating findings

Results

Four main themes emerged from the data: Unpreparedness; Communication; Role of Others; and Professional Discomfort, which was recognised as occurring across the three other themes (Figure 1).

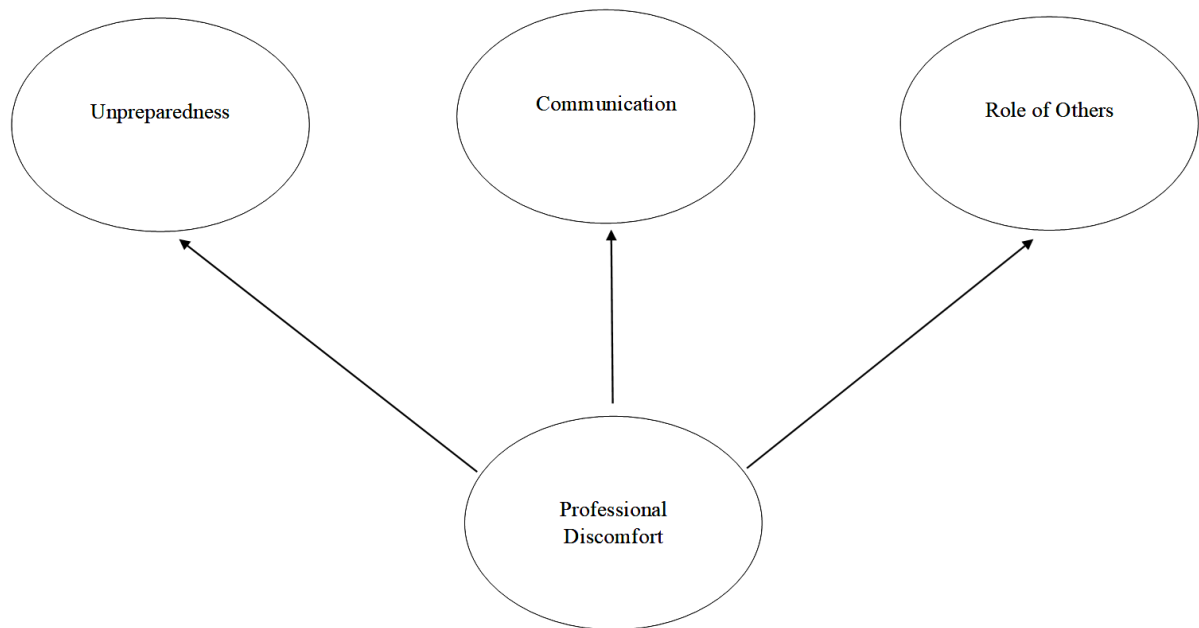


Figure 1. Map of themes and how they relate

Unpreparedness

This theme focuses on the sense of not feeling prepared to work with women with learning disabilities and was apparent across all six participant interviews. Four healthcare professionals reported that they received no education on working with people with learning disabilities during their training. For the two participants who had received teaching on working with patients with learning disabilities, this appeared to not be specific to people with learning disabilities, but rather on patient groups who may have other difficulties with

communication, such as those with sensory impairments or those who did not have English as their first language.

'It was more about a visual impairment...you know if people had hearing or if they were blind... it was more that, so I don't think we ever did if I really think about it. It was more of be aware throughout your training that there's lots of people with disabilities.' Deborah

'Lectures that were things from communicating with people who might struggle so deaf or blind people to learning difficulties and also people who maybe don't have English as their first language. I don't think we have that much just on learning disabilities.' Carol

The idea that education on working with people with learning disabilities is considered brief and appended onto something deemed more important was further demonstrated.

'I think it's briefly picked up on in study sessions for safeguarding... It's part of it but for me only a small part. It's more focused on- which it should be- paediatrics. I think we can certainly put more into it than they do.' Frances

Despite the described lack of sufficient teaching, it was felt by some participants that when they began their careers in clinical practice, they were expected to know how to effectively work with this patient group.

'I remember starting in 2012 new into general practice... not had any training and I was literally thrown in at the deep end... I remember my first learning disability patient. What do you say? What do you do? How do you talk to them?' Beatrice

The idea of feeling 'in at the deep end' and similar metaphors to explain their experience of feeling unprepared was echoed in other participant responses.

'...It's a new role for me, and I've kind of plunged into the deep end with it. It's quite a steep learning curve'. Carol

'It wasn't so much a training, more of a kind of ...I don't want to say baptism of fire, but you know we were basically...' Emma

Feeling unprepared to work with women with learning disabilities presenting clinically was contrasted by some participants with their experience of working with patients with dementia, and the resources available to help them work with that patient group.

'I think it would be good if there was some kind of thing out there...training like you can with dementia awareness' Carol.

Additionally, one participant highlighted that patient records did not always contain information about their learning disability, which they believed further hindered their sense of preparedness.

'To be honest, on her notes, there's no real clear documentation about it, which I thought was weird...about what type it is, what sort of communication skills you might need...there wasn't anything like that, it shocked me'. Anne

Communication

The theme of communication was evident across all six participant interviews. The intimate aspect of breast and cervical screening was considered a key motivator by the participants in ensuring that communication was effective, as was the complexity in explaining the purpose and ramifications of BRCA gene testing.

'I think mainly the concern is around communication, making sure the other person understands it, and giving all the information in the easiest way possible for them to understand it.' Anne

'I think they really need to understand... [BRCA testing] is difficult to get your head around even if you haven't got any learning disabilities. I think there's probably alternatives to how you relay that information...like a visual aid.' Deborah

The importance of the details of the cervical screening procedure being communicated to patients beforehand was discussed, but the fact that this was likely to be a standard letter and not adapted was reflected upon as being a potential barrier to women with learning disabilities accessing this screening.

'They receive a letter saying it's time for your smear, as far as I know it's just a generic letter that goes out, "phone your GP and book your appointment". If they've received a letter and they don't like the look of it, they could just put it in the bin' Carol.

Communication was perceived to be a key component in ensuring mammograms were less distressing for patients, particularly if the patient had not been informed of the purpose of the screening, or what the procedure would entail prior to the appointment. However, there appeared to be uncertainty that even with additional information that this would be understood by the female with a learning disability attending breast screening.

'I don't think they have an awareness of why they're here...they've don't know why the doctor's feeling that part of the breast or whatever which makes it difficult...I just try to talk to them and explain how I do with anybody, maybe slightly word it differently... as I said though, they might not understand that anyway...you've just got to be careful and word things as best you can really'. Frances

Participants perceived adapting communication to be an important aspect of ensuring that women with learning disabilities could feel included in their own care, but there was an awareness that the nature of professional systems were not always conducive to this.

'Because nursing can be quite technical and some of the terminology can be quite ridiculous'

Anne

'I just feel often in medicine we assume people know things and assume patients have a greater understanding than they do...There are resources out there, but it doesn't seem to be kind of just embedded'. Carol

'Some doctors do fire it, you know, very medically...People who haven't got learning disabilities, I've asked if they understood, have gone, "Well, no not really". They might not want to speak up...so they're going to do that even less with a learning disability'. Deborah

'I'd like to think that more time was given to those patients, definitely. However, I'm not convinced on that'. Emma

The recognition of needing to adapt communication appeared to be met with some uncertainty of how this would be received by the patient, and alluded to the professionals' own sense of potential uneasiness,

'If they're non-verbal I will still sit and talk to them because I'm not going to treat them any differently to a normal patient coming in the building' Beatrice

'I wouldn't want to patronise and say "oh, should I draw you a picture?"...I won't want to feel embarrassed to draw the picture for someone who's got learning disabilities if that's offensive to do it' Carol

The importance of communicating with the patient to establish their abilities and wishes was recognised as significant, particularly with regards to the impact that this may have on clinical decision making and healthcare provisions.

'Sometimes patients so much surprise you so much, they are very reserved and yet they have an absolute full understanding of what's going on, regardless of how they've been billed if that makes sense... I mean there's a wider issue, you know, should we be denying patients treatment based on what we think they're able to handle?' Emma

Role of Others

This theme encapsulates the perceived impact and importance of the role of others within the both the patients' and professionals' wider systems, which was apparent across all interviews. This theme was closely linked to 'Communication', in that both women with learning disabilities and the participants often needed support from others to help facilitate communication, however the 'Role of Others' was perceived to encompass other factors in addition to communication.

There appeared to be a consensus amongst the participants that they believed a female patient with a learning disability was unlikely to attend an appointment alone, and would invariably be accompanied by a parent or carer.

'I think because of the learning disability, the support is very in your face...They come as a package, with the carer'. Emma

The perception that parents or carers hold an important role in the patients' lives and hence their medical care was evident across all six interviews. Parents and carers were considered an important asset for healthcare professionals in undertaking procedures with women with learning disabilities, particularly with regards to helping the patient prepare for the appointment, and in supporting the healthcare professionals' information gathering during appointments.

'I think parents that have that conversation with them before they come to the appointment is really good because they're already ready for what's going to happen' Beatrice

'I think a collateral history is so important in situations like that, if the carers know her well'
Carol

'I think other things that can help is those people like the carers and families who know her really well but can give you sorts of tips how to manage the situation' Anne

'It does help to have a little bit of background information on what they understand and what they don't understand' Deborah

Some healthcare professionals perceived parents or carers as gatekeepers in the decision to perform screening for the female patients with a learning disability, but that this may be influenced by the parents' or carers' own assumptions and wishes. This seemed particularly evident in the case of cervical screening, and the potential erroneous association between this screening only being necessary for sexually active women.

'You've sent the letter out addressed to the patient with learning disabilities, parents are going to open it and go, 'she's not sexually active', and just chuck it in the bin...It's not a normal thing to kind of discuss with your child. It worries me actually that we're missing patients' Beatrice.

'To decide, for example, with a cervical screening because that's so intimate and whether that would be appropriate... we would have a chat with the family as to what they want'

Carol

The influential role of others in the patient's life was further reflected upon, particularly with regards to how health services may overlook patients that face challenges in seeking cancer screening independently, such as the female patients with a learning disability.

'I think patients are being missed, and I do wonder whether a lot more people have died of some form of cancer that's not been found because they've not been offered this screening.

How many doctors chase patients?' Beatrice

There appeared to be the perception that women with learning disabilities were in a powerless position, with parents', carers', and healthcare professionals' views instrumental in whether they were able to access screening services or not. All participants were keen to emphasise their awareness of their duty to provide equitable care to women with learning disabilities, but that equitable care in practice could be a fragile construct if others involved in the patient care were not mindful of this.

'I feel very strongly that our process is facilitating that, but I think that somebody can break the chain at any point with a comment or something that blocks the patient' Emma

The role of specialist staff was alluded to across the interviews. There seemed to be a sense that women with learning disabilities would have their needs better met by staff who were solely for them, and not general patients.

'I think a named learning disability nurse in the hospital would be amazing. I just think it would improve care so much and make it easy. I think to have them in different conversations around different cancer screenings' Beatrice

Professional Discomfort

The theme of professional discomfort appeared to be a fundamental undercurrent in all participant interviews, and to the themes of 'Unpreparedness', 'Communication', and 'Role of Others'. This theme is important as it encompasses the less overt perceptions of the

healthcare professionals towards providing cancer screening and BRCA gene testing to women with learning disabilities.

The sense of feeling unprepared conveyed from the participant interviews may indeed be as a result of feeling that they received insufficient teaching. However, this could also be suggestive of a less salient process of healthcare professionals deflecting the emotional discomfort they experience from feeling beyond their zone of comfort when working with females with a learning disability.

'I felt a lot of pressure, I was like, so really uncomfortable doing it anyway, because of, I could see she was quite distressed, but at the same time I could see where they were coming from.' Anne

'That can be a bit scary if you know that you've got someone coming in to discuss something, but you don't know them, if you've not met them before, I think.' Carol

'It was almost as if there was no interest and that's what was quite upsetting and quite moving really almost. Yeah, because like I say obviously the patient couldn't communicate at all for herself whatsoever.' Frances

Externalising this discomfort by placing responsibility with Universities or their employer may serve to ameliorate this feeling.

'I don't know, I might be wrong, but I think learning disabilities is seen as a bit of a taboo area...So yeah, I think universities let people down.' Beatrice

'There's nothing actually, that we've been given on how to relay this sort of information to patients that have got learning disabilities.' Anne

'I wish training could be better for staff overall...If the NHS can't get it right with training and awareness then what hope has society got? At the end of the day we are a caring profession-if we can't do it right who is going to do it right.' Frances

'I think it is a bit taboo in some ways as well because you don't have that much teaching'

Carol.

The perception of feeling unable to communicate with women with learning disabilities evidently led to feelings of discomfort for the professionals personally, but also demonstrated their concerns around the negative consequence this may have for the patient.

'The thing that I find really difficult is when you can't explain something to a patient for example needing to do an intimate exam and them getting quite upset...obviously you're doing it because it's in the patients best interest but it is really tricky when you don't know...just because someone can't communicate doesn't mean that they don't feel embarrassed' Carol

'What do you do with this patient that's got learning disabilities, can't consent, at what point do you continue a procedure that's too much for them...If they don't understand what a smear test is, it's probably going to cause a negative effect mentally for them'. Beatrice

'There's a fine line between telling them the truth...I guess it all depends on the understanding of the person as well and their emotional state, and the disability...someone might process it better and then be paranoid and fixated that they're going to get cancer and die from it.. I think that might be worse than not telling them' Frances

Within the theme of 'Role of Others', there appeared to be a sense of precariousness, with healthcare professionals sharing their concerns around the challenges faced by women with learning disabilities in accessing screening services. The perception that women with learning disabilities were powerless was conveyed, and the healthcare professionals appeared to find this an uncomfortable aspect of providing screening for this population. This was

evidenced by the concern that the patient's voice could be lost, or that they could be accompanied by somebody inappropriate, or receiving inadequate support.

'But then you've also got the issue of safeguarding, is that the right person to be with them, you know, because obviously there's other issues isn't there of questions and controlling. You have to be careful as well that those people don't speak for the individual' Deborah

'The carer didn't seem to you have any knowledge really. When it came to the point of giving her the physical examination obviously, she has to get undressed, take her top and bra off. It was filthy, yeah, quite upsetting. It was awful, I just found it really sad' Frances

There was a consensus from the healthcare professionals that further measures could be put in place to better support women with learning disabilities in accessing screening, but that these measures could possibly result in an increased workload for them, and paradoxically reduce the quality of care they provide.

'We're literally so busy' Beatrice

'I hesitate to say that more needs to be done... we all have a job to do on a very day-to-day basis...we have to keep up with everything, tick all the boxes if you like, in terms of keeping in touch with everyone for the right reasons at the right time... it might take us even further away from the actual patient rather than close to if you see what I mean' Emma.

There was also the perception that healthcare professionals felt that they needed to be providing ideal care, and that this could cause them to feel insecure within their roles when providing care to women with learning disabilities, due to feeling vulnerable to criticism.

'On a personal level I guess we need to be seen to be doing the right thing, without feeling that we are constantly going to be criticised for doing the wrong thing. Because we're so worried about that, you know'. Emma

'I'm one of these where I will walk away and say, nah, I'm not doing that. I'm not having a patient sitting there crying in front of me because they don't like what's going on'. Beatrice

Discussion

This research aimed to identify themes in the perceptions of healthcare professionals towards cancer screening and BRCA gene testing for females with a learning disability. Four themes emerged from the participant interviews: Unpreparedness, Communication, Role of Others, and Professional Discomfort. The latter appeared as a distinct theme, but also permeated the other themes. These themes will be discussed within the context of the wider existing literature and societal narratives. The clinical and research implications will also be discussed.

The theme of Unpreparedness emerged in part from the participants' reflections that they felt the teaching they had received during their training was insufficient in meeting their information needs. Furthermore, there was also evidence of insufficient information being provided post-qualification within clinical settings. The responses appeared to convey an awareness from the healthcare professionals that there may be nuances in working with people with learning disabilities, and thus categorising teaching on this specific patient group under an umbrella approach with other communication difficulties seemed inappropriate. The NHS Long Term Plan (2019) has committed to providing all NHS staff with education on the needs of people with learning disabilities to ensure that health services are supported to make reasonable adjustments, and the findings of the present study appear to support this as a welcome contribution to improve and consolidate skills and knowledge. The provision of education focused specifically on people with learning disabilities is of further importance when considering the historical and contemporary context of abuse and inequality

experienced by people with learning disabilities in healthcare services (Flynn & Citarella, 2013; Heslop et al., 2013; Mencap, 2007).

The themes of Communication and Role of Others can be understood within the context of the Social Model of Disability (Oliver, 2013). Participants' reflections regarding the inaccessibility of correspondence highlighted that health services may contribute to structural and psycho-emotional disability. The participants in this research recognised that adapting their own communication was necessary, but doing this in practice could be uncomfortable and hence deter them from doing so, which could possibly have the unintended consequence of further disabling the female patients with a learning disability. Furthermore, there were concerns that women with learning disabilities were at risk of not having their voice heard in the presence of parents or care staff, which is consistent with existing theories of power and intersectionality (Johnstone, 2018).

All themes alluded to the pressure that the healthcare professionals experienced in wanting to feel skilled and knowledgeable in working with women with learning disabilities, and the discomfort that arises when this does not feel achievable. The inherent nature of working in a caring profession, in which problems require attention and resolution through practical means, may exacerbate feelings of discomfort when healthcare professionals are faced with challenges that require a reflective approach. Feeling pressured to be the 'ideal' caregiver may further be reflective of the current zeitgeist in which healthcare professionals are presented within a 'hero narrative' or as the personification of 'angels' (Stokes-Parish, Elliot, Rolls & Massey, 2020). Existing research has highlighted that when working with patients with learning disabilities, better outcomes and improved patient satisfaction are achieved when the principal skills of empathy, listening and compassion are demonstrated (Tuffrey-Wijne, 2009). Feeling insecure within one's role may limit the capacity to engage with these skills, and this was discussed by the participants in the present study. Participation

in reflective practice focused on working with patients with learning disabilities has been demonstrated in previous research to be effective in developing healthcare professionals' confidence working with this patient group, during training and post-qualification (Fitzsimmons & Barr, 1997; Honey, Waterworth, Baker & Lenzie-Smith, 2006).

Strengths and Limitations

The current study brings attention to the lack of research and resources available regarding BRCA gene testing in females with learning disabilities and is a novel contribution to this subject area. The results also stimulate further discussions around improving communication and accessibility to cervical screening and breast screening, which is of significance when considering the low rates of participation in these screenings and higher mortality rates in females with learning disabilities.

Engagement in reflective discussions by the lead researcher and the research supervisor throughout the process of conducting, analysing and writing the research could also be considered a strength of the current study (Barker & Pistrang, 2005).

All participants in the study had experience of providing healthcare to people with learning disabilities, and hence their views may not be fully representative in respect to healthcare professionals without such experience.

Whilst the research highlights the apparent lack of voice afforded to women with learning disabilities, the study could have further benefited by including women with learning disabilities. For example, women with learning disabilities could have been consulted at different stages of the research, such in the initial stages of planning the project or in the creation of the questions included in the semi-structured interview.

Research and Clinical Implications

The emotional impact of providing cancer screening to females with learning disabilities experienced by healthcare professionals is an area that warrants further investigation. This appears to be especially relevant for BRCA gene testing, which can pose ethical challenges and dilemmas for healthcare professionals irrespective of whether the patient has a learning disability or not (Harris, Winship & Spriggs, 2005).

Clinician skill could be enhanced by supervision being provided by senior colleagues experienced in working with women with learning disabilities. Furthermore, the provision of reflective spaces, during training and post-qualification, to help healthcare professionals with the emotional impact of their role is likely to be beneficial for staff wellbeing and confidence in performing their role when providing care to women with learning disabilities, which consequently could improve the experience for the patients. Attendance at Schwartz Rounds, designed to offer supportive reflection to staff of different professional backgrounds, could also be encouraged to support this (Flanagan, Chadwick, Goodrich, Ford, & Wickens, 2020).

The importance of ensuring females with learning disabilities were prepared and informed of the procedures prior to their appointment was discussed by the participants, thus research focused on interventions to help facilitate this would be beneficial. This could be enhanced further by primary care and acute services linking up with learning disability services. As key stakeholders, women with learning disabilities should be included in the development of information resources around cancer screening procedures, and be consulted as experts by experience in training programmes for healthcare professionals during their training and post-qualification.

Furthermore, routine inclusion of specialist liaison staff should be encouraged, as this was identified by the participants as a helpful resource. Research including women with learning disabilities as participants focused on cancer screening and BRCA testing would

allow for their contribution and voice to be heard, which again was identified by the participants as an area of concern.

Conclusions

The findings of this study suggest that healthcare professionals can feel unprepared in providing cancer screening to female patients with learning disabilities. This appeared to be as a result of perceived insufficient education and discomfort associated with the emotional impact of the role. Issues around communication, accessibility, and the influential role of others were also discussed in relation to providing screening services, and healthcare professionals are likely to benefit from reflective practice to help with the emotional impact associated with their role. The recognition that the patient voices need to be heard is important and should be the focus of future research, in view of the healthcare disparities this population experiences.

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Part Three: Appendices

Appendix A: Submission Guidelines for the British Journal of Learning Disabilities

1. SUBMISSION

Authors should kindly note that submission implies that the content has not been published or submitted for publication elsewhere except as a brief abstract in the proceedings of a scientific meeting or symposium.

Once the submission materials have been prepared in accordance with the Author Guidelines, manuscripts should be submitted online at: <http://mc.manuscriptcentral.com/BLD>.

[Click here](#) for more details on how to use ScholarOne. Full instructions and support are available on the site and a user ID and password can be obtained on the first visit. If you require assistance then click Help link which appears at the top right of every Manuscript Central page.

If you cannot submit online, please contact the Editorial Office by email: BLDedoffice@wiley.com

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By submitting a manuscript to or reviewing for this publication, your name, email address, and affiliation, and other contact details the publication might require, will be used for the regular operations of the publication, including, when necessary, sharing with the publisher (Wiley) and partners for production and publication. The publication and the publisher recognize the importance of protecting the personal information collected from users in the operation of these services, and have practices in place to ensure that steps are taken to maintain the security, integrity, and privacy of the personal data collected and processed. You can learn more at <https://authorservices.wiley.com/statements/data-protection-policy.html>.

Preprint policy

Wiley believes that journals publishing for communities with established pre-print servers should allow authors to submit manuscripts which have already been made available on a non-commercial preprint server. Allowing submission does not, of course, guarantee that an article will be sent out for review. It simply reflects our belief that journals should not rule out reviewing a paper simply because it has already been available on a non-commercial server. Please see below for the specific policy language.

However, Wiley also knows that the use of preprint servers is not universally accepted and that individual journals and/or societies may approach submission of preprints differently.

This journal will consider for review articles previously available as preprints on non-commercial servers such as ArXiv, bioRxiv, psyArXiv, SocArXiv, engrXiv, etc. Authors may also post the submitted version of a manuscript to non-commercial servers at any time. Authors are requested to update any pre-publication versions with a link to the final published article.

2. AIMS AND SCOPE

The *British Journal of Learning Disabilities* is an interdisciplinary international peer-reviewed journal that draws contributions from a wide community of international researchers. It encompasses contemporary debate/s and developments in research, policy and practice that are relevant to the field of learning disabilities. Learning disabilities here refers to intellectual (global) disabilities and not to specific learning disabilities like dyslexia. The scope includes:

- activism and advocacy
- communication, interaction and relationships
- community lives and quality of life
- education and employment
- families and advocacy
- health and wellbeing
- policy, law and rights
- profound and multiple learning disabilities/additional needs

- research methods and inclusive research
- social care and professional roles.

The journal publishes original papers, commissioned keynote reviews on major topics, and book/resource reviews with some Special Issues giving comprehensive coverage to specific subject areas. The editor welcomes suggestions of topics for future Special Issues.

The journal supports dialogue with people with learning disabilities. *In Response* pieces provide feedback on the issues arising in the journal papers and their relevance for the lives of people with learning disabilities and those supporting them.

The international audience for *British Journal of Learning Disabilities* includes academics, professionals, practitioners, families and people with a personal and professional interest in learning disability. Authors are expected to consider this wide readership, to communicate in an accessible manner, and to exhibit knowledge of previously-published articles when submitting their work for consideration for publication. Careful attention to respectful terminology and ethical treatment is essential. You can see the journal's position on ethics [here](#).

The *British Journal of Learning Disabilities* is the official journal of the British Institute of Learning Disabilities. The opinions expressed in articles, whether editorials or otherwise, do not necessarily represent the official view of the British Institute of Learning Disabilities and the Institute accepts no responsibility for the quality of goods or services advertised.

3. MANUSCRIPT CATEGORIES AND REQUIREMENTS

- All Manuscripts submitted to *British Journal of Learning Disabilities* should include: Accessible Summary, Keywords, Abstract, Main Text (divide by appropriate sub headings) and References.
- Manuscripts should not be more than 7,000 words in length including references.

4. PREPARING THE SUBMISSION

Parts of the Manuscript

The manuscript should be submitted in separate files: title page; main text file; figures.

Title page

The title page should contain:

- A short informative title that contains the major key words. The title should not contain abbreviations (see Wiley's [best practice SEO tips](#));
- A short running title of less than 40 characters;
- The full names of the authors;
- The author's institutional affiliations where the work was conducted, with a footnote for the author's present address if different from where the work was conducted;
- Acknowledgments.

Abstract and Accessible Summary

Papers should include a structured abstract incorporating the following headings: Background, Methods, Findings, Conclusions. These should outline the questions investigated, the design, essential findings and main conclusions of the study. As well as an abstract, authors must include an easy-to-read summary of their papers. This was introduced in 2005, and was done so in the spirit of making research findings more accessible to people with learning disabilities. The editorial board also believe that this will make 'scanning' the Journal contents easier for all readers. Authors are required to:

- Summarise the content of their paper using bullet points (4 or 5 at most),

- Express their ideas in this summary using straightforward language, and
- State simply why the research is important, and should matter to people with learning disabilities.

Authorship

Please refer to the journal's authorship policy the Editorial Policies and Ethical Considerations section for details on eligibility for author listing.

Acknowledgments

Contributions from anyone who does not meet the criteria for authorship should be listed, with permission from the contributor, in an Acknowledgments section. Financial and material support should also be mentioned. Thanks to anonymous reviewers are not appropriate.

Conflict of Interest Statement

Authors will be asked to provide a conflict of interest statement during the submission process. For details on what to include in this section, see the section 'Conflict of Interest' in the Editorial Policies and Ethical Considerations section below. Submitting authors should ensure they liaise with all co-authors to confirm agreement with the final statement.

Main Text File

As papers are double-blind peer reviewed the main text file should not include any information that might identify the authors.

The main text file should be presented in the following order:

- i. Title, accessible summary, abstract and key words;
- ii. Main text;
- iii. References;
- iv. Tables (each table complete with title and footnotes);
- v. Figure legends;
- vi. Appendices (if relevant).

Figures and supporting information should be supplied as separate files.

Tables

Tables should be self-contained and complement, not duplicate, information contained in the text. They should be supplied as editable files, not pasted as images. Legends should be concise but comprehensive – the table, legend, and footnotes must be understandable without reference to the text. All abbreviations must be defined in footnotes. Footnote symbols: †, ‡, §, ¶, should be used (in that order) and *, **, *** should be reserved for P-values. Statistical measures such as SD or SEM should be identified in the headings.

Figure Legends

Legends should be concise but comprehensive – the figure and its legend must be understandable without reference to the text. Include definitions of any symbols used and define/explain all abbreviations and units of measurement.

Figures

Although authors are encouraged to send the highest-quality figures possible, for peer-review purposes, a wide variety of formats, sizes, and resolutions are accepted.

[Click here](#) for the basic figure requirements for figures submitted with manuscripts for initial peer review, as well as the more detailed post-acceptance figure requirements.

Color Figures. Figures submitted in color may be reproduced in colour online free of charge. Please note, however, that it is preferable that line figures (e.g. graphs and charts) are supplied in black and white so that they are legible if printed by a reader in black and white. If an author would prefer to have figures printed in colour in hard copies of the journal, a fee will be charged by the Publisher.

Data Citation

In recognition of the significance of data as an output of research effort, Wiley has endorsed the FORCE11 Data Citation Principles and is implementing a mandatory data citation policy. Wiley

journals require data to be cited in the same way as article, book, and web citations and authors are required to include data citations as part of their reference list. Data citation is appropriate for data held within institutional, subject focused, or more general data repositories. It is not intended to take the place of community standards such as in-line citation of GenBank accession codes. When citing or making claims based on data, authors must refer to the data at the relevant place in the manuscript text and in addition provide a formal citation in the reference list. We recommend the format proposed by the Joint Declaration of Data Citation Principles:

[dataset] Authors; Year; Dataset title; Data repository or archive; Version (if any); Persistent identifier (e.g. DOI)

References

References should be prepared according to the *Publication Manual of the American Psychological Association* (6th edition). This means in text citations should follow the author-date method whereby the author's last name and the year of publication for the source should appear in the text, for example, (Jones, 1998). The complete reference list should appear alphabetically by name at the end of the paper.

A sample of the most common entries in reference lists appears below. For more information about APA referencing style, please refer to the [APA FAQ](#). Note that for journal articles, issue numbers are not included unless each issue in the volume begins with page one, and a DOI should be provided for all references where available.

Journal article

Beers, S. R., & De Bellis, M. D. (2002). Neuropsychological function in children with maltreatment-related posttraumatic stress disorder. *The American Journal of Psychiatry*, 159, 483–486. doi:10.1176/appi.ajp.159.3.483

Book

Bradley-Johnson, S. (1994). *Psychoeducational assessment of students who are visually impaired or blind: Infancy through high school* (2nd ed.). Austin, TX: Pro-ed.

Internet Document

Norton, R. (2006, November 4). How to train a cat to operate a light switch [Video file]. Retrieved from <http://www.youtube.com/watch?v=Vja83KLQXZs>

Additional Files

Appendices

Appendices will be published after the references. For submission they should be supplied as separate files but referred to in the text.

Supporting Information

Supporting information is information that is not essential to the article, but provides greater depth and background. It is hosted online and appears without editing or typesetting. It may include tables, figures, videos, datasets, etc.

[Click here](#) for Wiley's FAQs on supporting information.

Note: if data, scripts, or other artefacts used to generate the analyses presented in the paper are available via a publicly available data repository, authors should include a reference to the location of the material within their paper.

General Style Points

The following points provide general advice on formatting and style.

- **Abbreviations:** All symbols and abbreviations should be clearly explained. Abbreviations should not be used when they refer to people (e.g. learning disabilities, not LD; developmental disabilities, not DD; intellectual disabilities, not ID)
- Please also use “people with learning disabilities” wherever possible, not “learning disabled people”.

- **Units of measurement:** Measurements should be given in SI or SI-derived units. Visit the [Bureau International des Poids et Mesures \(BIPM\) website](#) for more information about SI units.
- **Numbers:** numbers under 10 are spelt out, except for: measurements with a unit (8mmol/l); age (6 weeks old), or lists with other numbers (11 dogs, 9 cats, 4 gerbils).
- **Trade Names:** Chemical substances should be referred to by the generic name only. Trade names should not be used. Drugs should be referred to by their generic names. If proprietary drugs have been used in the study, refer to these by their generic name, mentioning the proprietary name and the name and location of the manufacturer in parentheses.

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Author Guidelines Updated September 2019

Appendix B: Papers rejected after whole paper was read

McEnhill, L. S. (2008). Breaking bad news of cancer to people with learning disabilities. *British Journal of Learning Disabilities*, 36(3), 157-164.

Tuffrey-Wijne, I. (2009). 'Am I a good girl?' Dying people who have a learning disability. *End of Life Care*, 3(1), 35-39.

**Appendix C: The National Institute for Health and Care Excellence Checklist for
Qualitative Studies**

Study identification: Include author, title, reference, year of publication		
Guidance topic:	Key research question/aim:	
Checklist completed by:		
Theoretical approach		
<p>1. Is a qualitative approach appropriate?</p> <p>For example:</p> <ul style="list-style-type: none"> • Does the research question seek to understand processes or structures, or illuminate subjective experiences or meanings? • Could a quantitative approach better have addressed the research question? 	<p>Appropriate</p> <p>Inappropriate</p> <p>Not sure</p>	<p>Comments:</p>
<p>2. Is the study clear in what it seeks to do?</p> <p>For example:</p> <ul style="list-style-type: none"> • Is the purpose of the study discussed – aims/objectives/research question/s? • Is there adequate/appropriate reference to the literature? • Are underpinning values/assumptions/theory discussed? 	<p>Clear</p> <p>Unclear</p> <p>Mixed</p>	<p>Comments:</p>
Study design		

<p>3. How defensible/rigorous is the research design/methodology?</p> <p>For example:</p> <ul style="list-style-type: none"> • Is the design appropriate to the research question? • Is a rationale given for using a qualitative approach? • Are there clear accounts of the rationale/justification for the sampling, data collection and data analysis techniques used? • Is the selection of cases/sampling strategy theoretically justified? 	<p>Defensible</p> <p>Indefensible</p> <p>Not sure</p>	<p>Comments:</p>
<p>Data collection</p>		
<p>4. How well was the data collection carried out?</p> <p>For example:</p> <ul style="list-style-type: none"> • Are the data collection methods clearly described? • Were the appropriate data collected to address the research question? • Was the data collection and record keeping systematic? 	<p>Appropriately</p> <p>Inappropriately</p> <p>Not sure/inadequately reported</p>	<p>Comments:</p>
<p>Trustworthiness</p>		
<p>5. Is the role of the researcher clearly described?</p> <p>For example:</p> <ul style="list-style-type: none"> • Has the relationship between the researcher and the participants been adequately considered? • Does the paper describe how the research was explained and presented to the participants? 	<p>Clearly described</p> <p>Unclear</p> <p>Not described</p>	<p>Comments:</p>

<p>6. Is the context clearly described?</p> <p>For example:</p> <ul style="list-style-type: none"> • Are the characteristics of the participants and settings clearly defined? • Were observations made in a sufficient variety of circumstances • Was context bias considered 	<p>Clear</p> <p>Unclear</p> <p>Not sure</p>	<p>Comments:</p>
<p>7. Were the methods reliable?</p> <p>For example:</p> <ul style="list-style-type: none"> • Was data collected by more than 1 method? • Is there justification for triangulation, or for not triangulating? • Do the methods investigate what they claim to? 	<p>Reliable</p> <p>Unreliable</p> <p>Not sure</p>	<p>Comments:</p>
<p>Analysis</p>		
<p>8. Is the data analysis sufficiently rigorous?</p> <p>For example:</p> <ul style="list-style-type: none"> • Is the procedure explicit – i.e. is it clear how the data was analysed to arrive at the results? • How systematic is the analysis, is the procedure reliable/dependable? • Is it clear how the themes and concepts were derived from the data? 	<p>Rigorous</p> <p>Not rigorous</p> <p>Not sure/not reported</p>	<p>Comments:</p>
<p>9. Is the data 'rich'?</p> <p>For example:</p> <ul style="list-style-type: none"> • How well are the contexts of the data described? 	<p>Rich</p> <p>Poor</p> <p>Not sure/not reported</p>	<p>Comments:</p>

<ul style="list-style-type: none"> • Has the diversity of perspective and content been explored? • How well has the detail and depth been demonstrated? • Are responses compared and contrasted across groups/sites? 		
<p>10. Is the analysis reliable?</p> <p>For example:</p> <ul style="list-style-type: none"> • Did more than 1 researcher theme and code transcripts/data? • If so, how were differences resolved? • Did participants feed back on the transcripts/data if possible and relevant? • Were negative/discrepant results addressed or ignored? 	<p>Reliable</p> <p>Unreliable</p> <p>Not sure/not reported</p>	<p>Comments:</p>
<p>11. Are the findings convincing?</p> <p>For example:</p> <ul style="list-style-type: none"> • Are the findings clearly presented? • Are the findings internally coherent? • Are extracts from the original data included? • Are the data appropriately referenced? • Is the reporting clear and coherent? 	<p>Convincing</p> <p>Not convincing</p> <p>Not sure</p>	<p>Comments:</p>
<p>12. Are the findings relevant to the aims of the study?</p>	<p>Relevant</p> <p>Irrelevant</p> <p>Partially relevant</p>	<p>Comments:</p>
<p>13. Conclusions</p>	<p>Adequate</p>	<p>Comments:</p>

<p>For example:</p> <ul style="list-style-type: none"> • How clear are the links between data, interpretation and conclusions? • Are the conclusions plausible and coherent? • Have alternative explanations been explored and discounted? • Does this enhance understanding of the research topic? • Are the implications of the research clearly defined? <p>Is there adequate discussion of any limitations encountered?</p>	<p>Inadequate</p> <p>Not sure</p>	
<p>Ethics</p>		
<p>14. How clear and coherent is the reporting of ethics?</p> <p>For example:</p> <ul style="list-style-type: none"> • Have ethical issues been taken into consideration? • Are they adequately discussed e.g. do they address consent and anonymity? • Have the consequences of the research been considered i.e. raising expectations, changing behaviour? • Was the study approved by an ethics committee? 	<p>Appropriate</p> <p>Inappropriate</p> <p>Not sure/not reported</p>	<p>Comments:</p>
<p>Overall assessment</p>		
<p>As far as can be ascertained from the paper, how well was the study conducted? (see guidance notes)</p>	<p>++</p> <p>+</p> <p>-</p>	<p>Comments:</p>

Appendix D: Quality assessment checklist ratings

Study	Theoretical Approach	Study Design	Data Collection	Validity and Reliability	Analysis	Ethics	Overall Assessment
Cresswell and Tuffrey-Wijne (2008)	Qualitative approach is appropriate	Single descriptive case study. No aims/objectives/research question discussed. No reference to literature or underpinning theory.	Not discussed	Personal account is likely to be valid, though not generalisable.	None	A note on ethical issues is presented, including the participant's decision and right to waive anonymity, as is the Research Steering Group's consideration and support of this. Ethical approval granted by the South East Multi Centre Ethics Committee	-
Flynn, Hubert-Williams, Hubert-Williams & Bramwell (2016) United Kingdom	Qualitative approach is appropriate	Qualitative study adopting semi-structured interviews is well designed and executed. Aims and objectives are clear, with good	Appropriate data collection to address the research question.	Only 6 participants were recruited into the study and interviews were relatively short, however participants were	Grounded theory is appropriate as the research aims to generate theories and hypotheses. The process of data analysis is well	Ethical approval granted by a University Departmental Ethics Committee and the North Wales (Central and East)	++

		reference to existing literature and theory.		demographically broad. The method and analysis appear to be sound.	documented. Direct quotations support analysis and conclusions are well presented. The primary researcher conducted the analysis with regular team/supervisory meetings as per best practice guidelines.	Research Ethics Committee. All participants were assessed as having capacity to give their informed consent to take part in the study. Pseudonyms were given to participants to ensure anonymity. Role of researcher clearly identified in relation to participants and their data. No conflicts of interest reported.	
Martean, Dallos, Stedmon & Moss (2013) United Kingdom	Qualitative approach is appropriate	Single case study adopting narrative analysis is well designed. Aims and objectives are clear, with good reference to	Appropriate data collection to address the research question.	The method and analysis appear to be sound, making reference to steps taken to ensure validity.	Analysis procedure explained in good detail. An interpretive panel to generate hypotheses and	Participant assigned a pseudonym to ensure anonymity. Informed consent for participating	++

		existing literature and theory.		Personal account is likely to be valid, though not generalisable.	produce different interpretative responses.	obtained using adapted materials. Full ethical approval granted by Local Research Ethics Committee, Bristol Royal Infirmary. Role of researcher clearly identified in relation to participants and their data	
Tuffrey-Wijne, Bernal, Jones, Butler & Hollins (2006) United Kingdom	Qualitative approach is appropriate	Aims and objectives are clear, with good reference to existing literature and theory.	Appropriate data collection to address the research question.	The method and analysis appear to be sound, making reference to steps taken to ensure validity. As a qualitative, hypothesis generating study, the method is clear that it does intend to generalise to a larger population.	Analysis described in good detail. Participants were active in verifying the results.	Ethical approval obtained from the South East Multi Centre Research Ethics Committee. Role of researcher clearly identified in relation to participants and their data	+

				Further limitations are given considerations. Resources used for data collection are of a good standard.			
Tuffrey-Wijne and Davies (2006) United Kingdom	Qualitative approach is appropriate	Single descriptive case study. Some reference to existing literature and theory.	Data collection for this individual paper is not discussed.	Personal account is likely to be valid, though not generalisable.	None	Ethical approval granted by the South East Multi Centre Ethics Committee. A Project Steering Group (including three members with a learning disability) helps to ensure that attention is continually focused on ethically sensitive issues.	-
Tuffrey-Wijne, Bernal, Hubert, Butler & Hollins (2009)	Qualitative approach is appropriate	Qualitative, ethnographic study adopting thematic field	Appropriate data collection to address the research question.	The method and analysis appear to be sound, making reference to steps	Grounded theory analysis described in good detail. Inclusion of	Ethical approval obtained from the South East Multi Centre Research	++

United Kingdom	analysis is well designed and executed. Aims and objectives are clear, with good reference to existing literature and theory.	taken to ensure validity. As a qualitative, hypothesis generating study, the method is clear that it does intend to generalise to a larger population.	NVivo 7 software to support data management.	Ethics Committee. Role of researcher clearly identified in relation to participants and their data			
Tuffrey-Wijne, Bernal & Hollins (2010) United Kingdom	Qualitative approach is appropriate	Qualitative, ethnographic study adopting thematic field analysis is well designed and executed. Aims and objectives are clear, with good reference to existing literature and theory.	Appropriate data collection to address the research question.	The method and analysis appear to be sound, making reference to steps taken to ensure validity. As a qualitative, hypothesis generating study, the method is clear that it does intend to generalise to a larger population. Researcher bias is considered and addressed by	Grounded theory analysis described in good detail. Inclusion of NVivo 7 software to support data management.	Ethical approval granted by multi-centre ethics committee. Ethical issues around participants' capacity to consent were consistently reviewed. Role of researcher clearly identified in relation to participants and their data	++

				external supervision from a psychotherapist			
Tuffrey-Wijne, Bernal, Hubert, Butler & Hollins (2010) United Kingdom	Qualitative approach is appropriate	Qualitative, ethnographic study adopting thematic field analysis is well designed and executed. Aims and objectives are clear, with good reference to existing literature and theory.	Appropriate data collection to address the research question.	The method and analysis appear to be sound, making reference to steps taken to ensure validity. As a qualitative, hypothesis generating study, the method is clear that it does intend to generalise to a larger population.	Grounded theory analysis described in good detail	Ethical approval granted by multi-centre ethics committee. Ethical issues around participants' capacity to consent were consistently reviewed. Role of researcher clearly identified in relation to participants and their data	++

Appendix E: Data extraction tool with worked example

Author, Title & Year of Publication	Flynn et al., 2016 “You don’t know what’s wrong with you”: an exploration of cancer related experiences in people with an intellectual disability
Location	United Kingdom
Research Aims	To better understand how people with a learning disability experience cancer, and generate theory and further research questions
Research Methodology	Design: Qualitative study Measures: Semi-structured interviews Analysis: Grounded theory
Ethics	All participants were assessed as having capacity to give their informed consent to take part in the study. Participants were provided with information that had been made accessible for them. Study approved by a University Departmental Ethics Committee and the North Wales (Central and East) Research Ethics Committee.
Participants	6 people (3 males, 3 females) with a learning disability and cancer aged over 18; and 12 further participants from their supportive network.
Summary of Findings	People with learning disabilities can be overlooked during their cancer care and feel excluded. Healthcare professionals often relied upon family members or care staff to facilitate communication, but healthcare professionals who included the patient mediated their distress associated with appointments. Cancer patients with learning disabilities were reluctant to share their distress with caregivers, this led to them appearing cut-off from their experience. Caregivers also appeared to withhold information

	<p>form the person with learning disabilities to protect them from psychological distress, however this was viewed negatively by the person with learning disability. Providing information to the patient facilitated feelings of empowerment.</p>
<p>Limitations</p>	<p>Relatively small sample size (however the author notes that theoretical saturation was reached). Only participants with a mild learning disability were recruited to the study (due to the requirement of participants to give informed consent), and the author recommends that a more heterogeneous sample would include cancer patients with more severe learning disabilities.</p> <p>Some limitations with richness of patient data that needed supplementing with caregiver or healthcare staff contributions.</p>
<p>Recommendations for Future Research</p>	<p>Importance of obtaining healthcare professionals' perspectives and experiences of providing care for this patient population in future research.</p> <p>Additional focus should also be on interventions designed to improve healthcare professionals' and care givers' understanding and knowledge of providing care to this patient group.</p> <p>Future research should be focused on the experience of having cancer in people with learning disabilities, particularly with regards to their experience of psychological distress.</p>

Appendix F: Semi-structured interview schedule

Interview Schedule

- 1. How long have you worked in the NHS for?
- 2. Can you tell me about your job role (main duties/responsibilities)
- 3. Have you received any training on working with patients with a learning disability? (when this was, how long it lasted, what was the content?)
- 4. Have you had any experience of providing care to females with a learning disability? (when this was, how long it lasted, what did this involve?)

If yes, these questions will be asked	If no, these questions will be asked
<ul style="list-style-type: none"> • 5. What was that experience like for you? Did you feel prepared/ confident in your abilities? 	<ul style="list-style-type: none"> • 5. If you have not any had any experience; how would you perceive this to be? How confident do you feel to do this?
<ul style="list-style-type: none"> • 6. Was it different to how you thought it would be? Where there any specific challenges or were there anything which supported you in this role? 	<ul style="list-style-type: none"> • 6. What do you think would be the specific challenges or facilitators to helping?
<ul style="list-style-type: none"> • 7. What do you think that experience was like for the patient (<i>ask them to think about anything they noticed, the person said, rather than just what they think the person experienced</i>) 	
<ul style="list-style-type: none"> • 8. Did you receive any feedback from the individual or possible carers? (What was this like? How did this impact on you? What was the nature of the feedback) 	

- 9. Have you ever had any experiences of providing support to women with a learning disability undergoing cancer screening?

If yes, these questions will be asked	If no, these questions will be asked
<ul style="list-style-type: none"> • 10. What was that like? Did you have any concerns prior to or during the experience? 	<ul style="list-style-type: none"> • 10. Would you have any concerns about this role if you were asked to support someone undergoing cancer screening?

<ul style="list-style-type: none"> 11. What do you think that was like for them? What things did you observe? Did the patient say anything to you? 	<ul style="list-style-type: none"> 11. What do you think the challenges/facilitators to this experience
<ul style="list-style-type: none"> 12. What do you think the challenges/facilitators to this experience were for you? 	

- 13. Have you ever had any experiences of providing support to women with a learning disability undergoing BRCA genetic testing or those identified as being at risk due to the gene?

If yes, these questions will be asked	If no, these questions will be asked
<ul style="list-style-type: none"> 14. What was that like for you? 	<ul style="list-style-type: none"> 14. Would you have any concerns about this role if you were asked to support someone undergoing BRCA genetic testing?
<ul style="list-style-type: none"> 15. What do you think that was like for the person with learning disabilities (i.e., did you think they had sufficient understanding, did they receive any accessible information? 	<ul style="list-style-type: none"> 15. What do you think the challenges/facilitators to this experience /would be?
<ul style="list-style-type: none"> 16. What do you think the challenges/facilitators to this experience were? 	

Appendix G: Recruitment e-mail

Are you a registered healthcare professional involved in patient cancer screening procedures? Would you like to take part in research exploring perceptions of providing care for female patients with a learning disability? Taking part in the research will involve a 1 hour interview and participants will be asked to answer questions about their perceptions of cancer screening and testing for the BRCA gene in females with a learning disability.

For further information or to register interest, please contact Taylor McAteer on 07***** or T.C.McAteer@2013.hull.ac.uk

Appendix H: Recruitment Poster for Social Media

IRAS ID: 241604
V. 1.2
18.06.2019



Volunteers needed for research study

Are you a healthcare professional?

Would you like to take part in research exploring perceptions of providing care for female patients with a learning disability?

Taking part in the research will involve a 1 hour interview and participants will be asked to answer questions about their perceptions about general cancer screening and testing for the BRCA gene in females with a learning disability.

For further information or to register interest, please contact Taylor McAteer on [REDACTED] or T.C.McAteer@2013.hull.ac.uk

Appendix I: Participant Information Sheet

INFORMATION SHEET FOR PARTICIPANTS

YOU WILL BE GIVEN A COPY OF THIS INFORMATION SHEET

Title of study Cancer screening and BRCA gene testing in females with a learning disability

I would like to invite you to participate in a research project which forms part of my doctorate research. Before you decide whether you want to take part, it is important for you to understand why the research is being done and what your participation will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask me if there is anything that is not clear or if you would like more information.

Experience of working with patients with a learning disability is not necessary.

What is the purpose of the study?

The purpose of the study is to explore the perceptions of healthcare professionals towards screening for cancer in females with learning disabilities. Existing research has identified that individuals with learning disabilities are more likely to experience inequalities in healthcare, and this is especially true for females with a learning disability. It is therefore of interest to gain healthcare professionals' perspectives on this.

What will happen if I take part?

If you agree to take part, I will ask you to answer some questions about your professional background. Then you will have a semi-structured interview with me which will last around 60 minutes. I will audio record the discussion. There are no right or wrong answers, and I am only interested in your opinions, your beliefs and your experiences of cancer screening for females with a learning disability.

Do I have to take part?

Participation is completely voluntary, and you should only take part if you want to. Once you have read this information sheet, please contact me if you have any questions that will help you make a decision about taking part. If you decide to take part, I will ask you to sign a consent form and you will be given a copy of this consent form to keep.

What are the possible risks of taking part?

Participating in the study will require 60 minutes of your time and this may be inconvenient for you. The discussed content may be upsetting for some, and it is recommended that you arrange a meeting with your own professional supervisor or the Occupational Health department of your organisation if you require support.

What are the possible benefits of taking part?

We cannot promise that you will have any direct benefits from taking part in the study. However, it is hoped that the information you give us will help us to understand more about the experiences of healthcare professionals when providing healthcare to females with learning disabilities. Sometimes people find it useful to have the opportunity to talk about their experiences.

Data handling and confidentiality

Your data will be processed in accordance with the General Data Protection Regulation 2018 (GDPR).

All of the personal information that you provide will be kept strictly confidential. Any information that could be used to identify you will not be used in the research. Direct quotes from the discussion may be used in research publications and presentations but you will not be identified in these. To protect your anonymity you will be assigned a code or pseudonym. This will ensure it will not be possible to identify you from the information you provide. To protect the security of the audio recordings an encrypted recording device will be used. After the research is completed, all of the audio recordings will be destroyed. Anonymised transcripts of the recordings will be stored securely in an on-line storage repository at the University of Hull for a period of ten years. The only time that information cannot be kept confidential is if you disclose something that suggests that you or someone else is at risk of serious harm. If this happens during the interview the researcher will need to contact appropriate authorities to ensure that you and other people are safe. It is unlikely that this will happen and the researcher will try to discuss this with you.

Your contact details will be held securely for the duration of the research but then destroyed when the research is complete.

Data Protection Statement

The data controller for this project will be the University of Hull. The University will process your personal data for the purpose of the research outlined above. The legal basis for processing your personal data for research purposes under GDPR is a 'task in the public interest' You can provide your consent for the use of your personal data in this study by completing the consent form that has been provided to you. Information about how the University of Hull processes your data can be found in the Research Privacy notice which will be given to you.

You have the right to access information held about you. Your right of access can be exercised in accordance with the General Data Protection Regulation. You also have other rights including rights of correction, erasure, objection, and data portability. Questions, comments and requests about your personal data can also be sent to the University of Hull Information Compliance Manager, Mr Luke Thompson (l.thompson3@hull.ac.uk). If you wish to lodge a complaint with the Information Commissioner's Office, please visit www.ico.org.uk.

What if I change my mind about taking part?

You are free to withdraw at any point of the study, without having to give a reason. Withdrawing from the study will not affect you in any way. You are able to withdraw your data from the study up until data analysis has commenced, after which withdrawal of your data will no longer be possible as the data will have been anonymised and/or committed to the final report. If you choose to withdraw from the study before this point the data collected will be destroyed.

What will happen to the results of the study?

The results of the study will be summarised in a written thesis as part of a Doctorate in Clinical Psychology. The thesis will be available on the University of Hull's on-line repository <https://hydra.hull.ac.uk/>. The research may also be published in academic journals or presented at conferences.

If you would like to receive a copy of the report once the study has been completed, please sign the appropriate section on the consent form indicating you would like to provide me with your contact details, which will be securely stored.

Who should I contact for further information?

If you have any questions or require more information about this study, please contact me using the following contact details:

Taylor McAteer

Email address: T.C.McAteer@2013.hull.ac.uk

What if I have further questions, or if something goes wrong?

If you wish to make a complaint about the conduct of the study, you can contact the University of Hull using the research supervisor's details below for further advice and information:

Dr Emma Lewis

Clinical Psychology

Aire Building

The University of Hull

Cottingham Road

Hull

HU6 7RX

Tel: +44 (0) 1482 464617

Email address: e.lewis@hull.ac.uk

Thank you for reading this information sheet and for considering taking part in this research.

Appendix J: Participant Consent Form

CONSENT FORM

Title of study: Cancer screening and BRCA gene testing in females with a learning disability

Name of Researcher: Taylor McAteer

Please
initial box

1. I confirm that I have read the information sheet dated 18/06/2019 (version 1.3) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my legal rights being affected, up until data analysis has commenced, after which withdrawal of my data will not be possible.
3. I understand that the research interview will be audio recorded and that my anonymised verbatim quotes may be used in research reports and conference presentations.
4. I give permission for the collection and use of my data to answer the research question in this study.
5. I agree to take part in the above study.
6. Please indicate if you would like to provide your contact details for the researcher to send you a copy of the final report once the study has been completed (please circle your response) **YES / NO**

If you have circled YES please provide your details:

Contact number:

Email address:

Name of Participant:

Date:

Signature:

Name of Person
taking consent:

Date:

Signature:

Appendix K: Ethical Approval Letters

Removed for digital archiving.

Removed for digital archiving.

Appendix L: Participant Sources of Support

Thank you for taking part in the study. Please retain this leaflet.

Cancer is a sensitive topic to discuss, and some people may find that talking about cancer is difficult or upsetting for them. If you have become distressed or upset as a result of participating in this study, please discuss this with your line manager, clinical supervisor, or contact your Trust's Occupational Health Department.

Further sources of information and support can be accessed via Macmillan's website www.macmillan.org.uk

Or by calling [0808 808 00 00](tel:08088080000) (lines are 7 days a week, 8am-8pm)

Local support is available from the following services

Macmillan Cancer Information and Support Service Hull

Tel: 01482 461154

The Queen's Centre For Oncology And
Haematology,
Castle Hill Hospital,
Castle Road, Hull
HU16 5JQ

Goole Information & Support Service

Tel: 03033 305374

Goole and District Hospital

Macmillan Drop-in session 10am-12
midday every Friday
Outpatients Department
Woodland Avenue
Goole
DN14 6RX

Scunthorpe Information and Support Service

Tel: 03033 305372

Macmillan Information Centre Scunthorpe,
Church Lane Entrance
Scunthorpe General Hospital, Cliff Gardens
Scunthorpe, North Lincolnshire
DN15 7BH

Grimsby Information & Support Service

Tel: 03033 305374

Macmillan Information & Support Service,
Amethyst Suite
Diana, Princess Of Wales Hospital,
Scartho Road,
Grimsby, North East Lincolnshire
DN33 2BA

**For urgent confidential support please contact the Samaritans on
116 123. Lines are free and monitored 24/7**

Appendix M: Worked Example of Data Analysis

Transcript	Initial Codes	Emergent Theme
<p>Carol: There haven't been any new residents in the care home, so I haven't had to set this up, but we would then have a best interest meeting.</p> <p>To decide, for example, with a cervical screening because that's so intimate. And whether that would be appropriate. So, 98% of cancer, cervical cancers are caused by the HPV virus, which you get through sexual activity, so if...if the women that have never had sex then that greatly reduces the risk of a cervical cancer. And then we have a chat with family as to what they want, if they...if they can't communicate or a chat with the patient. There was only that one, but it was in...in the age range to still have smears that one lady...</p> <p>...but we didn't do them for her. She did have a referral a couple of years ago because she had a very heavy bleeding.</p> <p>But...so then when she went to gynae, they had a look at her...her cervix looked healthy and then we just decided we should go on symptoms rather than to having to put her through a smear that we can't explain to her properly.</p> <p>Researcher: Sure. Yeah...yeah, okay.</p> <p>Carol: And but the other patients then with the faecal testing...that's quite easy because like the carers can...can help them with that or just do it for them.</p>	<p>Making decisions based on best interests</p> <p>Making assumptions about patient lifestyle</p> <p>Speaking to family</p> <p>Physical symptoms present</p> <p>Differences in care provided</p> <p>Perceived challenges in communication</p> <p>Carer involvement</p>	<p>Professionals as guardians</p> <p>Assumptions</p> <p>Communication Role of others in patient's wider system</p> <p>Reactive care</p> <p>Protection</p> <p>Communication</p> <p>Role of others in patient's wider system</p>

<p>Researcher: Yeah...yeah. Okay. And that's done at home?</p> <p>Carol: Yes. So that's a little poo test that you take the sample of and then put in the post.</p> <p>Researcher: Okay. So, the next question then, have you ever had any experiences of providing support to women with a learning disability undergoing BRCA gene testing or those identified as being at risk due to the gene?</p> <p>Carol: I haven't, no. Not...not with learning difficulties, no.</p> <p>Researcher: Okay. So, if you did... if this did come up, would you have any concerns about this role if you were asked to support somebody undergoing BRCA, the genetic testing procedure?</p> <p>Carol: I don't think so. I...I feel really comfortable having tough conversations with people, and I feel like I can gauge the level of understanding quite well with people, so I would...if I was going to have to initiate a conversation like that, I would have a chat first and say things like, do you know what you've come for today? How much do you understand about what's going on? Do you want me to explain anything to you? And I try and go about it that way because I just feel like often in medicine, we assume people know things. And that means that things are missed out. We assume patients have a greater understanding that they do, but then, equally, I don't want to be someone to patronise</p>	<p>Establishing patient abilities and understanding</p> <p>Making assumptions</p> <p>Aware of the need to clarify level of understanding and ability but not wanting to appear patronising</p>	<p>Communication</p> <p>Assumptions</p> <p>Communication</p> <p>Professional discomfort</p>
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Appendix N: Reflective Statement

Prior to starting the course, I was aware that my research topic was likely to be focused on an aspect of physical health. I have always been interested in the psychological element of health and illness, and especially cancer. It was not until the research fair that I had considered focusing on cancer in people with learning disabilities. I was surprised at how little research there appeared to be when I initially started searching online, and the research that was available appeared to be from a small pool of authors. This appealed to my inner curiosity and I decided that this was a subject area I wanted to know more about, and hopefully contribute to through my own research.

During my clinical training I was fortunate enough to have two excellent placements in learning disability services. Having met individuals with learning disabilities and their families I was struck by how often I heard similar stories of their health complaints being dismissed as 'behavioural' or medical professionals not appearing able to provide an adequate amount of care, resulting in them feeling 'given up on'. It became clear that this was a broad issue, but that for my research I was going to need something more focused. I think I was drawn to the idea of BRCA gene testing, and breast and cervical screening in females with a learning disability as a result of my clinical experiences, and how important these procedures are in ensuring that any malignancies are detected early.

The Research Journey

For some time, I rather erroneously perceived the research aspect of the doctorate course to be a 'hoop to jump through'. I soon realised that this was not the case (or if it was, that I am not much of a hoop jumper). Throughout my life I have been very fortunate in being able to grasp ideas and concepts and get things done with relative ease. However, I found the thought of completing a thesis incredibly daunting, and this feeling permeated into actually coming

up with a question, and in the setting up of the research. My initial preference would have been to have women with learning disabilities directly involved in this research. I think it is important for their voices to be heard and I am somewhat regretful that my research was not able to do this, partly due to my own concerns regarding obtaining ethical approval around such a sensitive topic. I am aware that my research further adds to the body of literature about people with learning disabilities but not including them, and in the future I would endeavour to design research with people with learning disabilities as active participants in it.

I found the process of obtaining ethical approval particularly challenging; on reflection, I perceived the feedback provided to me as criticism and proof that I was not good enough to conduct research. In response to this I became somewhat avoidant. I experienced feelings of insecurity and frustration that the other trainees in my cohort appeared to be navigating the research systems with, what appeared to me, relative ease. Perhaps unsurprisingly, my confidence was affected by this comparison with others. It occurred to me that I had probably taken my ability to understand and get things done in the past for granted. This made me consider what it must be like to not be in such a position of privilege within society, and to regularly experience feelings of frustration and insecurity in navigating systems that pose less of a challenge to others.

A turning point came for me when I began my research interviews. I had some difficulties in recruiting participants, and this was further inhibited by not being able to go into the research sites I had spent so much time trying to gain access to due to COVID-19 restrictions. The discussions I had with the healthcare professionals who kindly took part in my research helped me further develop my confidence in my project, and my understanding of it too.

I found myself identifying with the idea of being 'in at the deep end' that was conveyed by the participants of this research. Being relatively young and inexperienced prior to

commencing my Clinical Psychology training, at times I have felt unprepared and found myself quarrelling with self-doubt. I have also been aware of the pull to 'over-care' and feeling under pressure to provide 'perfect' care to those that I work with clinically. Completing this research has challenged my ideas around perfectionism and made me appreciate being 'good enough', and I am sure that this lesson will affect other aspects of my career and life positively.

Final Thoughts

The Systematic Literature Review and Empirical paper presented in my portfolio thesis both demonstrate the need for high quality and equitable care at all stages of the cancer experience for people with learning disabilities. Recent events have brought social inequalities to the public's attention and there appears to be a greater awareness and motivation for individuals and organisations to make changes to address these. I hope that this extends into healthcare too. It was reassuring to find that the healthcare professionals who took part in my research were keen to know more and were motivated to provide high quality care for this patient group.

Reading about people's cancer experiences had a special poignancy for me, having lost relatives to this disease; one of whom passed away during my first year of the doctorate. The impact of poor health, especially a cancer diagnosis, on an individual's wellbeing is something that I feel all people should be offered support with, regardless of ability.

Throughout the process of reviewing the literature, I noticed that the subject of cancer in learning disabilities can be somewhat taboo, and hence not really spoken about. I was struck by a phrase I heard during teaching: 'Discomfort offers fertile learning ground'. As a psychologist, the feeling of discomfort is something that I have become familiar and well-practised with. I was interested to discover the theme of discomfort emerging from the

participants in my empirical study, and how they may feel alone with this. I contrasted this to the privileged position of psychologists who are able to seek supervision or peer support about how they feel, and not just on what they do.

Having completed this research, I would like to think that I can use the skills that I have developed to conduct further research in the future. I am especially interested in further focusing on healthcare experiences in people with learning disabilities, as I found that there is a relatively thin body of literature out there. I hope my present contribution to this subject area is meaningful in some way. Whilst I have always been aware of the importance of research in informing practice, the process of conducting and writing up my own research has made me fully appreciate the value of research in shaping what we know and how we know it.

The process of completing a Doctorate in Clinical Psychology has been challenging and at times felt overwhelming, but overall, it is something that has ultimately changed my life for the better. I will always be grateful of the opportunity provided to me by the University of Hull and Humber Teaching NHS Foundation Trust.

Appendix O: Epistemological Statement

Epistemology is the theory of knowledge, encapsulating the nature, origin, and limits of human knowledge (Carter & Little 2007). It refers not only to what we know, but how we know what we know (Crotty, 1998). It is important to understand the epistemological stance of the researcher as this informs the approach and assumptions underpinning the research.

The researcher was aware that their experiences in learning disability services and psychological medicine services could have affected their own perceptions of this subject area. To minimise bias, themes were reviewed and discussed in research supervision, and reviewed by an independent professional. The researcher was keen to approach this research from an *a posteriori* position, given that they were from a different professional background than the participants in this research, but remained aware of their role in ‘authoring’ rather ‘discovering’ the knowledge generated from the research (Willig 2008). The aim of the research was to identify themes in the participants’ perceptions of cancer screening and BRCA gene testing in females with learning disabilities. Perception has been defined by Munhall (2012) as an ‘individual’s access to experience and interpretation’, and that one’s own perception can be understood through the lenses in which reality is viewed. As such, there is no one true ‘reality’, but rather multiple realities (Madill, Jordan & Shirley, 2000).

Thematic analysis was chosen due the theoretical and epistemological flexibility it offers (Braun & Clarke, 2006), and its usefulness in examining the perspectives of different research participants and allowing unanticipated findings to emerge (King, 2004).

Understanding perception from a position of critical realism, and wanting to obtain the perceptions of others, an inductive approach to thematic analysis was adopted. This approach allowed for themes to emerge from the data (Patton, 1990) rather than attempting to fit the data within an existing frame or in the researcher’s preconceptions (Braun & Clarke, 2006).

Further, the analysis was conducted at a latent level, which enabled the analysis to go beyond

the semantic level to allow further exploration of the assumptions underlying the data (Braun & Clarke, 2006).

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