Understanding Inequalities in Cancer Screening in the Learning Disability and Autism Populations of the Northern Isles

A Thesis Submitted in Partial Fulfilment of the Requirements for the Degree of
Professional Doctorate in Health Psychology

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Abstract

Background: Cancer screening programmes reduce the incidence of, and mortality from, cancer through early detection and intervention at a population level. People with learning disabilities and/or autism throughout the UK face considerable inequalities in cancer screening, and the Scottish Government have pledged to improve access to screening services for these populations. Research naturally focuses on the more urban populations which does not necessarily apply or translate to Scotland's vast remote and rural geography. The Northern Isles of Orkney and Shetland are on the periphery and provide unique challenges to population-based interventions and healthcare research. This thesis aims to better understand inequalities of access to cancer screening programmes for the learning disabled and autistic populations in Orkney and Shetland.

Methods: A clinical audit was conducted on available GP Practice data for cancer screening and HPV immunisation in the learning disability and autism populations of Orkney and Shetland. This was followed by a qualitative exploration of stakeholders' views on and experiences of cancer screening for people with learning disabilities and/or autism. Interviews and focus groups were analysed to develop themes using Reflexive Thematic Analysis.

Results: Rates of screening uptake are comparable to available data on the learning disabilities populations elsewhere in Scotland but serve to highlight a greater disparity in Orkney and Shetland, given the relatively high uptake of screening in their respective general populations. Participants described high-level structural barriers to screening, including screening systems and processes, uncertainty around responsibilities and decision-making, and how cancer and cancer screening is viewed in the community. Small communities were seen as a unique strength but were also felt to be a factor in perpetuating unhelpful myths and stereotypes.

Conclusion: Targeted efforts are essential to redress these inequities, for which we now have evidence-based recommendations and a co-production plan.

Keywords: *Learning Disabilities; Cancer Screening; HPV Immunisation; Scotland; Remote and Rural; Clinical Audit; Reflexive Thematic Analysis*

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Finally, I dedicate this thesis to my sister, Kaela, for being my port in a storm.

Declaration

I declare that, except where explicit reference is made to the contribution of others, this thesis embodies the results of my own research and was composed by me.

This thesis has not been submitted for any other degree at the University of Stirling or any other institution.

Signature:

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List of Acronyms and Abbreviations

CHI	Community Health Index
DNA	Did not attend
FIT	Faecal immunochemical test
GGC	Greater Glasgow and Clyde
GP	General Practitioner
HPV	Human Papilloma Virus
MDT	Multi-Disciplinary Team
NHS	National Health Service
PHS	Public Health Scotland
PI	Primary Investigator
RA	Research Assistant
SCIE	Social Care Institute of Excellence
SCLD	Scottish Commission for Learning Disabilities
SCCRS	Scottish Cervical Cytology Results System
SU	Service User
TA	Thematic Analysis
WHO	World Health Organisation
WwLD	Women with Learning Disabilities

Chapter 1. Introduction

Cancer screening programmes reduce the incidence of, and mortality from, cancer through early detection and intervention at a population level (World Health Organisation [WHO], 2020). Unfortunately, the reported uptake of cancer screening programmes in people with learning disabilities remains lower than that of the general population across the United Kingdom (Gray et al. 2018; McCowan et al., 2019; National Health Service [NHS] Digital, 2022; Osborn et al 2012). People with learning disabilities and/or autism face considerable health inequalities with increased risk of developing cancer, or physical health conditions which may in turn lead to cancer (Crepsi, 2011; Simantov et al., 2022; Weir et al., 2021). The formative publication on the principles and practice of screening by Wilson & Jungner (1968) specified that screening programmes must be acceptable to the population, as well as highlighting the necessity of education through accessible and appropriate formats, to ensure awareness and understanding of the benefits of screening. Over ten years have passed since the Equality Act 2010 made law that healthcare providers have a duty to ensure reasonable adjustments are made for people with learning disabilities and autism to receive equitable access to healthcare. Equitable screening for the learning disability and autistic populations remains aspirational and demands serious attention.

1.1 Overview of Cancer Screening and HPV Immunisation in Scotland

Scotland currently offers three cancer screening programmes, to which individuals are invited by letter, based on their age and/or sex.

1.1.1 Colorectal (Bowel) Cancer Screening

Bowel screening is offered to both men and women aged 50-74, every two years. Those aged 75 and above can continue to request tests. Those eligible are sent a faecal immunochemical test (FIT) to complete at home. The FIT test requires one small sample of faeces which is used to detect micrograms of human haemoglobin per gram of faeces, for which a positive result can be indicative of changes in the bowel. Should a positive result be found, the individual is invited for a colonoscopy to assess whether the bowel changes are due to cancer.

1.1.2 Breast Cancer Screening

Breast screening is offered every three years, between the ages of 50 and 70, to women, non-binary people assigned female at birth who haven't undergone breast removal, trans women who are taking hormones and trans men who haven't had breast removal surgery. Self-referral is also accepted for those aged 71-74, and those who have previously had breast cancer. Those eligible are invited to attend an appointment at one of six screening centres, or via a mobile screening unit for more remote and rural areas.

1.1.3 Cervical Cancer Screening

Cervical screening looks for the presence of Human Papilloma Virus (HPV) in the cervix. Almost all cervical cancers are caused by HPV (Xing et al., 2021). Women and anyone with a cervix aged 25-64 are invited to arrange a cervical screening appointment at their local GP Practice. The frequency of cervical screening changed in 2020, from every three years, to every five years. This change was due to the move from cytology to primary HPV testing, which has greater sensitivity and accuracy in detecting those at higher risk of cervical cancer (Rebolj et al., 2019). If changes are found which require monitoring, screening can be offered up to the age of 70.

1.1.4 HPV Immunisation

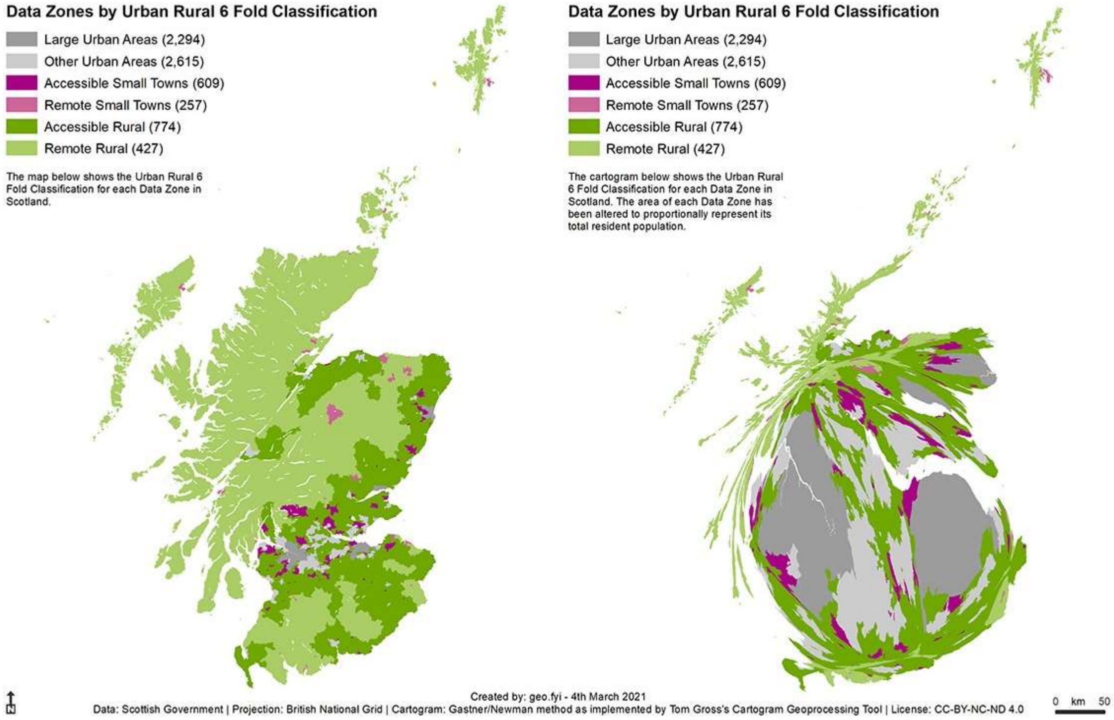
The introduction of the HPV vaccine in 2008, which is offered in secondary schools to all S1 pupils aged 12-13, has significantly reduced preinvasive cervical disease (Palmer et al., 2019). The vaccine has been found to be effective against the five types of HPV which account for 84% of invasive cervical cancers (Arbyn et al., 2014, Kavanagh et al. 2017).

1.2 Remote and Rural Contexts for Cancer Screening

In Scotland, the Scottish Government have pledged to improve access to screening services for people with learning disabilities and autism in policy, through funding projects and evaluating and sharing examples of best practice across the country (Scottish Government, 2019). Research naturally focuses on the more densely populated central belt, however, conclusions or recommendations drawn from samples in these urban populations do not necessarily apply or translate to Scotland's vast remote and rural geography, which contains pockets of small, self-contained and culturally diverse communities. These remote and rural populations amount to only 6% of the total population, but are spread across 70% of the total land mass (Scottish Government, 2021). Figure 1 provides a visual comparison of Scotland's geography with a cartogram altered to proportionally represent the populations, which emphasizes how population density could distort a sense of geography. Of the most remote areas of Scotland, the Northern Isles of Orkney and Shetland are on the periphery and provide unique challenges to population-based interventions and healthcare research, particularly for minority populations such as those with learning disabilities or autism.

Figure 2

A Visual Depiction of Scotland's Urban, Small Town and Rural Populations by Data Zone (2011) on a Standard Map and Proportionally Altered Cartogram.



Created by: geo.fyi - 4th March 2021
Data: Scottish Government | Projection: British National Grid | Cartogram: Gastner/Newman method as implemented by Tom Gross's Cartogram Geoprocessing Tool | License: CC-BY-NC-ND 4.0

Note: Source https://geo.fyi/2021/03/04/scotlands-data-zones/. Copyright geo.fyi. Used with permission.

1.3 Challenges in Data Collection

National statistics on the learning disability and autistic population of Scotland have to date been produced by the Scottish Commission for People with Learning Disabilities (SCLD) and Public Health Scotland (PHS). Data are based on individuals known to local authorities, but do not necessarily access services. Significant challenges exist in gathering accurate data on specific health outcomes of learning disability populations, including the lack of consistent or sufficiently disaggregated data (Gilling & Phillips, 2017; Primary Care Informatics, 2015) and delays to linking and sharing data between existing sources (Callander, 2021; SCLD, 2022). Existing data shows both Orkney and Shetland perform better than Scotland as a whole in the uptake of all three screening programmes and HPV immunisation. However, these figures may not necessarily translate to the learning disability and autism populations and to date no exploration has been made of the current status of cancer screening/HPV uptake in these populations in the northern isles.

1.4 Overview of the thesis

This thesis comprises two studies which aim to better understand inequalities of access to cancer screening programmes for learning disabled/autistic populations in Shetland and Orkney and covers three research questions:

- What does the local data tell us about cancer screening and HPV immunisation in the learning disability and autistic populations of Orkney and Shetland?
- What are stakeholders’ views on and experiences of cancer screening and HPV immunisation?
- How can we improve experiences, and promote the uptake of cancer screening and HPV immunisation programmes for the learning disability and autism populations of Orkney and Shetland?

Chapter two of this thesis seeks to answer the first research question through a clinical audit of available data on cancer screening and HPV immunisation in Orkney and Shetland. The audit provides context from which to explore the second research question, detailed in Chapter three: a qualitative investigation into stakeholders’ views and experiences of cancer screening and immunisation programmes in Orkney and Shetland using Braun and Clarke's Reflexive Thematic Analysis (Reflexive TA). The meaning generated from the qualitative study will deepen our understanding of the Shetland and Orkney contexts and the barriers to accessing screening, as well as highlighting any aspects of good practice. Chapter four concludes with recommendations for practice, a co-production plan to implement these recommendations, reflections on the research process, and recommendations for future research.

1.5 Caveats and Considerations

Some considerations are worth noting from the outset. Firstly, while learning disabled and autistic people are unique and diverse populations, they are considered together in terms of service provision due to overlapping historical diagnostic criteria (Thurm et al., 2019), a degree of co-occurring prevalence (Mackay et al., 2017) as well as shared challenges such as stigma and barriers to healthcare (Scottish Government, 2021). As such, the current thesis will consider the issues of cancer screening for both populations together, whilst being mindful that there are likely to be both commonalities and differences.

Second, both Shetland and Orkney's learning disability and autism populations are small, as are the sample sizes for the qualitative study. There are also significant gaps in the available data. For these reasons, comparative analysis between the Islands were not conducted. In addition, the small samples and small communities present challenges in preserving anonymity. The qualitative data from each Island group is not considered separately and area of work and gender are not identified where necessary to protect identities.

Finally, the qualitative chapter of the current thesis requires an acknowledgement of the researcher's position in relation to the research (Braun and Clarke, 2019). The researcher is local to Shetland and has worked within the NHS for nine years, including projects to reduce health inequalities in the learning disability population. The current research is part of a government-funded project to reduce health inequalities in cancer screening and is being submitted as partial fulfilment of the researcher's professional doctorate training in health psychology. As such, this project was considered from a health psychology perspective. These factors have all influenced the development and analysis of the current research.

Chapter 2. An Audit of Orkney and Shetland's Primary Care Demographic and Cancer Screening Data

2.1 Introduction

The remote island populations of Orkney and Shetland present a unique context to delivering population screening programmes, beyond the complexities of coordinating relevant stakeholders and agreeing protocols and pathways with individual health boards. The breast screening service, for example, operates from six screening centres across Scotland. Orkney and Shetland fall into the North-East Scotland area covered by the screening centre in Aberdeen. Mobile units visit the isles every three years and are based in the main towns (Kirkwall and Lerwick) for several weeks. Residents in the outer isles will take one or even two ferries, and travel up to a six-hour round trip to attend a screening appointment. Investigative colonoscopies following a positive bowel screening result would likewise require travel to the main towns. For cervical screening individuals are required to visit their registered GP Practice. Shetland's Primary Care comprises of ten GP Practices across the isles, and Orkney's Primary Care has six GP Practices. Smaller, remote islands are usually linked to the closest Practice and by visiting Nurse Practitioners. Despite these geographical challenges, Orkney and Shetland have some of the highest cancer screening uptake rates in Scotland.

2.1.1 Current Uptake of Cancer Screening and HPV Immunisation in the General Population

The screening and HPV immunisation programmes are nationally commissioned services, with each programme run through centralised systems, collating data through independent data systems. The data from cancer screening and HPV immunisation programmes in the general population in Scotland detailed below is published annually by Public Health Scotland (PHS, 2023). A summary of the most recent available uptake data for each screening programme across localities is presented in Table 1. Bowel screening uptake across Scotland between 2020 and 2022 was 66.7% (64.3% for males, 69.1% for females). The uptake of bowel screening in Orkney was 70.1% overall (67.2% for males, 73.0% for females) and for Shetland 74.3% overall (71.8% for males and 76.9% for females). For breast screening, the uptake within the three-year rolling period 2019/20 to 2021/2022 was 74.5% across Scotland. In Orkney, 83.6% of women participated in breast screening across this time period and in Shetland the rate was 85.2%. For cervical screening, the overall national rate of uptake as of 31st of March 2022 was 68.7%. Uptake has been in decline since 2018. As at this time, those aged 25-49 were screened every three years and those aged 49-64 were screened every five years, different calculations were used for older and younger women. Nationally, 65.7% of younger women participated in cervical screening, whereas 75.9% of this age group participated in Orkney, and 79.1% participated in Shetland. For women aged 50-64, the uptake nationally rose to 73.7%, but was still greater in Orkney at 76.3% and Shetland at 79.0%.

Table 1

Rates of Uptake of Cancer Screening Programmes in Orkney, Shetland and Scotland Overall (Public Health Scotland, 2023)

Screening Programme	Orkney	Shetland	Scotland
Bowel Screening[a]			
Total	70.1%	74.3%	66.7%
Males	67.2%	71.8%	64.3%
Females	73.0%	76.9%	69.1%
Breast Screening[b]	83.6%	85.2%	74.5%
Cervical Screening[c]			
Younger[d]	75.9%	79.1%	65.7%
Older[e]	76.3%	79.0%	73.7%

[a] Between 2020 and 2022; [b] Within the three-year rolling period 2018/2019 and 2020/2021; [c] Rate of uptake as of 31st of March, 2022; [d] 25-49 age group; [e] 50-64 age group

The routine HPV immunisation schedule consists of two doses of vaccine for full coverage, given in the first and second years of high school. Nationally, for the school year 2021/2022, 83.6% of S2 pupils were covered by the first dose (86.4% of females, 80.9% of males) and 61.6% were covered by the second dose (64.8% of females and 58.6% of males). For Orkney, the rate of coverage by the first dose was 84.4% of S2 pupils (86.8% of females and 82.4% of males) dropping to 73% of coverage by the second dose (77.4% of females and 69.5% of males). For Shetland, 90.1% of S2 pupils were covered by the first dose (89.9% of females and 90.3% of males), dropping to 79.6% of coverage by the second dose (77% of females and 82.1% of males). An overview can be seen in Table 2.

Table 2

HPV Immunisation Uptake for School Year 2021/2022 (Public Health Scotland, 2023)

Dose by Sex	Orkney	Shetland	Scotland
First dose			
Total	84.4%	90.1%	83.6%
Males	82.4%	90.3%	80.9%
Females	86.8%	89.9%	86.4%
Second dose			
Total	73%	79.6%	61.6%
Males	69.5%	82.1%	58.6%
Females	77.4%	77%	64.8%

It is important to note that the above data covers the time period of the Covid-19 pandemic, which reduced the uptake of all screening and immunisation as screening programmes in Scotland were temporarily paused.

2.1.2 Available Statistics on the Learning Disability and Autistic Population

Challenges in collecting data and building a picture of cancer screening uptake in the learning disability and autism populations are compounded by barriers to data sharing between the local authorities and NHS Boards. NHS Boards are dependent on data from Primary Care GP Practices for information on the learning disability and autism populations. However, individuals are no longer routinely coded as having a learning disability or autism, which presents difficulties for identifying the population and gaining an accurate understanding of screening uptake. Local authorities collect population data, but little health information. In addition, the population data collected by the local authorities are not necessarily analogous with the data collected by GP Practices.

Statistics on learning disability and autism population size are based on local authority data published by the Scottish Commission for People with Learning Disabilities (SCLD). The most recent available reports 23,584 people with learning disabilities living in Scotland, equating to 5.2 per 1000 in the general population, and 4383 adults identifying as being on the autism spectrum (SCLD, 2019). Shetland reported a known population of 164 individuals with a Learning Disability, equating to 8.7 per 1000 in the general population. This is the second highest of the 30 local authorities in Scotland who provided data, below Dundee City Council at 8.8 per 1000. Orkney's known Learning Disability population was seventh-lowest in Scotland at 91, equating to 4.9 per 1000 in the general population (SCLD, 2019).

Data published by NHS Digital on the coverage of cancer screening programmes in England has shown that the proportion of people with learning disabilities and/or autism who participate in the breast, bowel and cervical screening programmes is much lower than in the general population (NHS Digital, 2022). To date, there has been little investigation into the rate of uptake of cancer screening and HPV immunisation in the Learning Disability and/or Autism populations of Scotland. Research is now underway to rectify this and establish a profile of the health, mortality and health inequalities for the learning disability and autism population of Scotland (Cooper et al., 2022). Indications are that screening uptake in Scotland's learning disabilities and autism populations show a similar pattern to England. A retrospective cohort study by Osborn et al. (2012) using The Health Improvement Network: a primary care research database proportionately representative of the UK population, found significantly lower rates of uptake in all screening programmes in people with learning disabilities than the general population. More recently, data from a Scottish Health Board, NHS Greater Glasgow and Clyde (NHS GGC, 2022), show people with learning disabilities had poorer uptake in bowel, breast and cervical screening than the general population.

Little data is available on how factors such as age, or gender may relate to cancer screening uptake in the learning disabilities populations. The above data show a general trend of slightly lower uptake of bowel cancer screening and HPV immunisations in males both in the Isles and nation-wide. McCowan et al., (2019) found that increasing age and affluence in women in Glasgow was associated with higher cancer screening programme uptake. A few studies have looked at patterns in screening behaviours in the general population across programmes. Data from the Netherlands (Kregting et al., 2022), Denmark (Njor et al.,2023) and Scotland (McCowan et al., 2019) all found that just over 50% of women attended all three screening programmes. Whether these trends are mirrored in the learning disability or autism populations remains to be seen.

2.1.3 Aim and Audit Questions

The existing data on the learning disability and autism populations of Orkney and Shetland has to date never been extracted or compared with local authority data. An audit was decided upon in order to compile the data and enable cross-referencing with cancer screening uptake data. The aim of this audit is to better understand participation in cancer screening in the learning disability and autism populations of Orkney and Shetland, identify any patterns or gaps in the data available and make recommendations for improvement.

The primary data analysis sought to answer the following questions:

- How does the data compare to the national data on the learning disability/autism population participation in cancer screening, and on population data known to the local authority?
- What are the demographics of the populations?
- What proportion of Shetland and Orkney's populations recorded as having a Learning Disability or Autism are participating in cancer screening (breast, cervical and bowel), and the HPV immunisation programme?
- Are there any differences between those who participate or do not participate in terms of gender, age, and locality?
- What accessibility adjustments or improvements are made during appointments?
- How consistent is the data recording, and what (if any) gaps exist?

Secondary analysis will be conducted to ascertain whether women who participate in one form of screening are more likely to participate in others, and whether those who participate or do not participate in their first invitation are more or less likely to participate in future invitations.

2.2 Methods

Ethical approval was sought and granted by the NHS, Invasive or Clinical Research Panel of the University of Stirling on 4th October 2021 (appendix 1). Approval was also sought from both Health Boards respective Caldicott Guardians and Clinical Governance Teams in accordance with NHS Scotland protocol.

The audit was a retrospective review of all registered patients' records in all GP Practices in Orkney and Shetland. Patients were identified who had been recorded as having a Learning Disability or Autism through the EMIS system, used for patient records in Primary Care throughout Scotland. Patients were also identified where they have specific Learning Disability Administration codes recorded on their files. These records gave a total number of patients recorded as having a Learning Disability and/or

Autism in Shetland and Orkney. From this selected population, the Community Health Index (CHI) number and following basic demographic data was extracted: age, sex and health centre. In addition, information relating to specific clinical codes recorded against each individual indicating any accessibility or adjustments made during appointments, was noted. The data was extracted from the EMIS system by NHS Information Analysts with the appropriate permissions to access Primary Care data. The Information Analysts then used the CHI numbers to request cancer screening data from the relevant sources, before passing the collated data on to the principle investigator (PI; AJ). Data was collected between November 2021 and February 2023. Upon obtaining the complete dataset, the PI cleaned the data and transferred to a spreadsheet using IBM SPSS Statistics version 28 for analysis.

Data relating to the cervical screening and bowel screening programme was obtained from the Scottish Cervical Cytology Results System (SCCRS) and DISCOVERY database respectively. Data on breast cancer screening was obtained via central screening services in Grampian.

Bowel screening data included the dates of invitation and whether or not an individual attended. Breast screening data included the date of most recent screening where available, and the dates of any screenings not attended, or “DNAs”. Cervical screening included the date of most recent screening where available, dates when an individual was coded as a “defaulter” (failed to attend) and if and when they were ever coded as excluded from screening. Brief notes detailing the reason for exclusion were also available.

In addition to the screening data, each health centre was also asked what procedures they follow to make adjustments for accessibility and how these are recorded. Emails were sent to each health centre’s generic inbox, addressed to the Practice Manager, or were sent directly to the Practice Manager where known, asking for responses to the following questions:

- What, if any, procedures are in place within your Practice to record or register patients as having a learning disability and/or autism and requiring reasonable adjustments?

- What procedures are in place within your Practice to make reasonable adjustments for people with learning disabilities and/or autism?
- Are accessibility improvements or adjustments during appointments recorded? If so, how?
- What, if any, follow-up processes are in place within your Practice for defaulters/non-responders to the bowel, breast and cervical screening programmes?
- Do you have any other comments in relation to cancer screening for the learning disability and/or autism population?

The questions were open text but responses were expected to be brief. Practices were given three weeks to respond, before being prompted again via email. Where no response was received, a further prompt was issued. Responses were collated and examined using content analysis, and a coding framework developed to analyse the results.

2.3 Results

2.3.1 Overall Population

A total of 327 individuals were identified in fifteen GP Practices across Orkney and Shetland, having been coded as having a learning disability and/or autism. Data will not be discussed at the Practice level in order to preserve anonymity. One hundred and eighty of the individuals reside in Shetland (55%) and 147 in Orkney (45%), 16 more in Shetland and 56 more in Orkney than reported by the respective local authorities. The sample comprised of 203 males (62.1%) and 124 females (37.9%). There was a noticeable gender disparity in Shetland, with a 67.2% male population. An overview of the population characteristics and eligibility for screening is shown in Table 3.

The population age ranged from 1 to 88, with a mean age of 37.25. Eligibility for bowel and breast screening was defined as all those over the age of 50, and eligibility for cervical screening was defined as women over the age of 25. Those beyond the upper age limit of eligibility were included in the datasets, as their data could still contribute to analysis. A total of 75 individuals (22.9%) were eligible (or were previously eligible) for bowel screening; 29 (23.4%) women were over 50 eligible (or were previously eligible) for invitation to breast screening, 98 (79.0%) would have been eligible for invitation to cervical screening and 31 (9.5%) should have been offered the HPV vaccine by the time of data analysis.

Table 3

Overview of Population and Eligibility for Screening/HPV Immunisation

Population Characteristic		Orkney (n=147)	Shetland (n= 180)	Total (n=327)
Sex	Male	82 (55.8%)[a]	121 (67.2%)[a]	203 (62.1%)[a]
	Female	65 (44.2%)[a]	59 (32.8%)[a]	124 (37.9%)[a]
	Total	147 (45%)[b]	180 (55%)[b]	327
Rurality	Central	91 (61.9%)[a]	102 (56.7%)[a]	193 (59%)[b]
	Outskirt	56 (38.0%)[a]	78 (43.3%)[a]	134 (41%)[b]
Bowel screening eligibility	Male	22 (51.2%)[c]	24 (75%)[c]	46 (61.3%)[c]
	Female	21 (48.8%)[c]	8 (25%)[c]	29 (38.7%)[c]
	Total	43 (29.3%)[a]	32 (17.8%)[a]	75 (22.9%)[b]
Breast screening eligibility		21 (32.3%)[d]	8 (13.6%)[d]	29 (23.4%)[d]
Cervical screening eligibility		53 (81.5%)[d]	45 (76.3%)[d]	98 (79.0%)[d]
HPV vaccine eligibility	Male	2 (2.4%)[c]	12 (66.7%)[c]	14 (6.9%)[d]
	Female	11 (84.6%)[c]	6 (33.3%)[c]	17 (13.7%)[d]
	Total	13 (8.8%)[a]	18 (10.0%)[a]	31 (9.5%)[b]

Note: Eligibility based on age.

[a]Percentage of column total; [b]Percentage of grand total; [c]Percentage of total eligible within column; [d]Percentage eligible within total sex in column;

Table 4 shows individuals' residence by council electoral districts. These were categorised as 'central' (central mainland) or 'outskirts' (remote mainland/outer isles) to explore whether those living in more rural locations differed in their screening participation. Maps of Orkney and Shetland's electoral districts are provided in appendices 2 and 3, respectively.

Table 4

Number of Individuals With Learning Disability/Autism by Council Electoral Districts

Island Group	Council Electoral District	*n* of Population
Shetland		
	Shetland Central[a]	35 (19.4%)
	Lerwick North & Bressay[a]	25 (13.9%)
	Lerwick South[a]	42 (23.3%)
	North Isles	19 (10.6%)
	Shetland North	27 (15.0%)
	Shetland South	17 (9.4%
	Shetland West	15 (8.3%)
Orkney		
	East Mainland, South Ronaldsay & Burray	14 (4.3%)
	Kirkwall East[a]	45 (13.8%)
	Kirkwall West & Orphir[a]	46 (14.1%)
	North Isles	12 (3.7%)
	Stromness & South Isles	13 (4%)
	West Mainland	17 (5.2%)

Note. [a]Electoral districts categorized as 'central'; the remainder were categorized as 'outskirts'.

2.3.2 Bowel Screening

Of the 75 individuals in the dataset who were eligible for bowel screening at the time of analysis 43 (58.1%) lived in Orkney, and 32 (42.7%) lived in Shetland, 29 (39.2%) were female and 46 (60.8%) were male. The gender split was more even in Orkney, at 48.8% females and 51.2% in males, whereas in Shetland there were three times as many males (75%) than females (25%). Age ranged from 50-88. From the dataset it was possible to see how many invitations an individual had been issued and whether or not they ever participated. It was therefore possible to count how many screenings they participated in, and if there was any pattern. Six screening patterns were identified: always (participated upon every invitation); delayed (did not participate initially but subsequently began participating); intermittent (showed irregular patterns of participating and not); lapsed (began by participating in every invitation then ceased); yet to participate; and undefined (received only one invitation, no pattern to establish). Of the 75 eligible, seven had only just come of age, and two individuals had missing data. These individuals are not included in analysis, leaving a total of 66 individuals. An overview of these individuals' data can be seen in Table 5.

Table 5.

Overview of Bowel Screening Participation

Bowel Screening Participation		Island Board		Sex		Totals
		Orkney (n = 36)	Shetland (n = 30)	Male (n = 39)	Female (n = 27)	(n = 66)
Ever participated	Yes	16 (44.4%)	18 (60%)	21 (53.8%)	13 (48.1%)	38 (57.6%)
	No	20 (55.6%)	12 (40%)	18 (46.2%)	14 (51.9%)	28 (42.4%)
First screen	Yes	13 (36.1%)	14 (46.7%)	14 (35.9%)	13 (48.1%)	27 (40.9%)
	No	23 (63.9%)	16 (53.3%)	25 (64.1%)	14 (51.9%)	39 (59.1%)
Patterns of screening	Always	6 (16.7%)	11 (36.7%)	9 (23.1%)	8 (29.6%)	17 (25.8%)
	Delayed	4 (11.1%)	4 (13.3%)	5 (12.8%)	3 (11.1%)	8 (12.1%)
	Intermittent	4 (11.1%)	1 (3.3%)	3 (7.7%)	2 (7.4%)	5 (7.6%)
	Lapsed	2 (5.6%)	1 (3.3%)	1 (2.6%)	2 (7.4%)	3 (4.5%)
	Yet to participate	14 (38.9%)	12 (40%)	18 (46.2%)	8 (29.6%)	26[a] (39.4%)
	Undefined	6 (16.7%)	1 (3.3%)	3 (7.7%)	4 (14.8%)	7 (10.6%)

Note. % within columns

[a]Two individuals who have only been invited to one screening in which they did not participate, may still do so in future, so are recorded as "undefined" in terms of patterns of screening participation.

Thirty-eight (57.6%) of eligible individuals were recorded as having participated in bowel screening at least once, with 28 (42.4%) recorded as never having participated. A majority of 39 (59.1%) individuals did not return their first bowel screening test, compared to 27 (40.9%) who did. In terms of patterns of screening, individuals who have yet to participate in bowel screening make up the largest majority of 39.4% (n=26), whereas only 17 individuals (25.8%) were recorded as having returned every screening test. Due to the unequal gender split resulting in fewer older females in the Shetland dataset, a Mann-Whitney U test was run to determine if there were any statistically significant differences in age between males and females overall. Distributions of age were similar as assessed by visual inspection. Median age score was not statistically significantly different between males (median age = 61) and females (median age = 65), *U* = 12107, *z* = -.577, *p* = .564.

A binomial logistic regression was performed to ascertain whether island of residence, age, gender or rurality predicted having ever participated in bowel screening or not (Table 6). All assumptions were met. The logistic regression model was not statistically significant, $X^2(4) = 4.894$, *p* = .298.

Table 6.

Logistic Regression Predicting Likelihood of Bowel Screening based on Age, Gender, Island of Residence and Rurality

	B	*SE*	*Wald*	*df*	*p*	*Odds Ratio*	*95% CI for Odds Ratio*	
							Lower	Upper
Age	.009	.030	.090	1	.764	1.009	.952	1.070
Gender	.017	.526	.001	1	.975	1.017	.363	2.852
Island Board	.925	.514	3.247	1	.072	2.523	.922	6.904
Rurality	-.600	.576	1.085	1	.298	.549	.177	1.698
Constant	-.947	1.940	.238	1	.626	.388		

Other patterns of screening included: intermittent (n=5, 6.7%); lapsed (n=3, 4%); and delayed (n=8, 10.7%). Given the small numbers in each group it was not possible to conduct statistical analysis beyond these descriptives.

2.3.3 Breast Screening

Twenty-nine women in the dataset were eligible for breast screening. Age ranged from 50-79 (M=64.28). Of these, 21 (72.4%) were residing in Orkney, and only 8 (27.6%) resided in Shetland. Nineteen (65.5%) had participated in breast screening at least once, 6 (20.7%) were recorded as never having participated and 4 (13.8%) were missing data. An overview of the data can be seen in Table 7. DNA history, where participants had been invited but did not attend, was scant and not provided by any of the Orkney Practices.

Table 7.

Overview of Breast Screening Data

Participation in breast screening		Island Board		
		Orkney (n = 21)	Shetland (n = 8)	Total (n = 29)
Ever participated	Yes	16 (76.2%)	3 (37.5%)	19 (65.5%)
	No	1 (4.8%)	5 (62.5%)	6 (20.7%)
	Unknown	4 (19%)	0 (0%)	4 (13.8%)

Small numbers mean in-depth analysis was not possible beyond descriptive statistics. Living in the outskirts of the isles did not appear to influence participation in breast screening, as all 6 women recorded as non-responders lived in more central locations, and the 5 women living in more remote localities had all participated.

2.3.4 Cervical Screening

A total of 98 women were eligible for invitation to cervical screening, 53 (54.1%) residing in Orkney, and 45 (45.9%) residing in Shetland. Table 8 provides a summary of the data. Across both Boards, the majority were recorded as never having participated in cervical screening (53; 54.1%), with only 36 (36.7%) having participated at least once. Information was missing for 9 (9.2%) individuals. For Shetland, 29 (67.4%) individuals had never participated, against 14 (32.6%) who had, whereas in Orkney the split was more even, with 24 (52.2%) never participating and 22 (47.8%) participating at least once. These figures are mirrored in the recorded defaulter data (i.e. those who do not attend), but more individuals in Shetland are defaulting multiple times (18; 41.9%) than in Orkney (8; 17.4%). This may be explained by differences in recording; more individuals in Orkney are recorded as excluded from screening (26; 56.5%) compared to Shetland (10; 23.3%).

Rurality did not appear to show any pattern of influence on cervical screening behaviour, with proportions of individuals having participated in cervical screening compared to those who had not as similar in both central (41.2% vs 58.8%) and outskirt (39.5% vs 60.5%) localities.

Data on whether individuals participated in their first cervical screening was not available to ascertain if participating in the first invite increased participation in subsequent invites. It was also not possible to ascertain the timing of exclusions and whether attempts were made to try for screening prior to this, however of the 36 who participated at some point, 7 (19.4%) were also recorded as excluded and 24 (66.7%) had defaulted, 8 (22.2%) of whom had done so more than once.

Brief details were available for some exclusions: anatomically impossible (n=2); no cervix (n=3); opted out (n=12); not clinically appropriate (n=26); no further recall (n=8); co-morbidity (n=3); pregnancy (n=1); transferred out of Scotland (n=2).

Table 8.

Overview of Cervical Screening Participation

		Orkney (n = 53)	Shetland (n = 45)	Total (n = 98)
Ever participated	Yes	22 (47.8%)	14 (32.6%)	36 (36.7%)
	No	24 (52.2%)	29 (67.4%)	53 (54.1%)
Defaulter	Yes	25 (47.2%)	30 (66.7%)	55 (56.1%)
	No	21 (39.6%)	13 (28.9%)	34 (34.7%)
Multiple defaulter		8 (17.4%)	18 (41.9%)	26 (29.2%)
Excluded from screening		26 (56.5%)	10 (23.3%)	36 (40.4%)
Missing		7 (13.2%)	2 (4.4%)	9 (9.2%)

2.3.5 HPV Immunisations

Thirty-one individuals had been eligible for HPV immunisation at the time of analysis (see Table 9). Of these, 18 (58.1%) resided in Shetland and 13 (41.9%) resided in Orkney, 17 were female (54.8%) and 14 (45.2%) were male. Thirteen individuals (41.9%) were recorded as having received the vaccines (fully vaccinated), 9 (29%) had not and 9 (29%) were unknown. Eight of the unknown were residents of Orkney, so this may be due to a data recording issue, as 0 were recorded as not having the vaccine. Five (38.5%) individuals in Orkney and 8 (44.4%) in Shetland were recorded as receiving the vaccine. Of the 17 females, 7 (41.2%) had received the vaccine, with 3 (17.6%) recorded as not having received the vaccine, and 7 (41.2%) unknown, and for the 14 males, 6 (42.9%) had received the vaccine, 6 (42.9%) had not and 2 (14.3%) were unknown.

Table 9.

Overview of HPV Immunisation Uptake

		Orkney (n = 13)	Shetland (n = 18)	Male (n = 14)	Female (n = 17)	Total (n = 31)
Received	Yes	5 (38.5%)	8 (44.4%)	6 (42.9%)	7 (41.2%)	13 (41.9%)
	No	0 (0%)	9 (50%)	6 (42.9%)	3 (17.6%)	9 (29%)
	Unknown	8 (61.5%)	1 (5.6%)	2 (14.3%)	7 (41.2%)	9 (29%)

Rurality did not appear to show any influence over acceptance of the HPV vaccination, with 7 (58.3%) individuals residing in more rural addresses receiving the vaccine compared to 3 (25%) who did not, and 6 (31.6%) living in more central areas receiving the vaccine compared to 6 (31.6%) who did not. Of the remaining unknown cases, 7 of these were registered to more central residences, and 2 to more rural. Again, small numbers prevented further analysis.

2.3.6 Individuals eligible for all three screening programmes

Sixteen women were eligible for all three screening programmes at the same time, 10 (62.5%) from Orkney and 6 (37.5%) from Shetland. Fourteen resided in more central areas (87.5%) and 2 (12.5%) in more remote areas. While numbers are too low to draw confident conclusions, there is a potential pattern of three distinct groups of screening uptake (see Table 10). Five women (31.3%) who had only ever participated in one screening programme (*either* breast (n=1), bowel (n=2) or cervical (n=2)); 5 women (31.3%) who participated in bowel *and* breast only and 4 women (25%) who participated in cervical *and* breast only. Only one woman (6.3%) had participated in all three, and only one did not participate in any.

Table 10

Pattern of Screening Participation by Women Eligible for all Three Screening Programmes

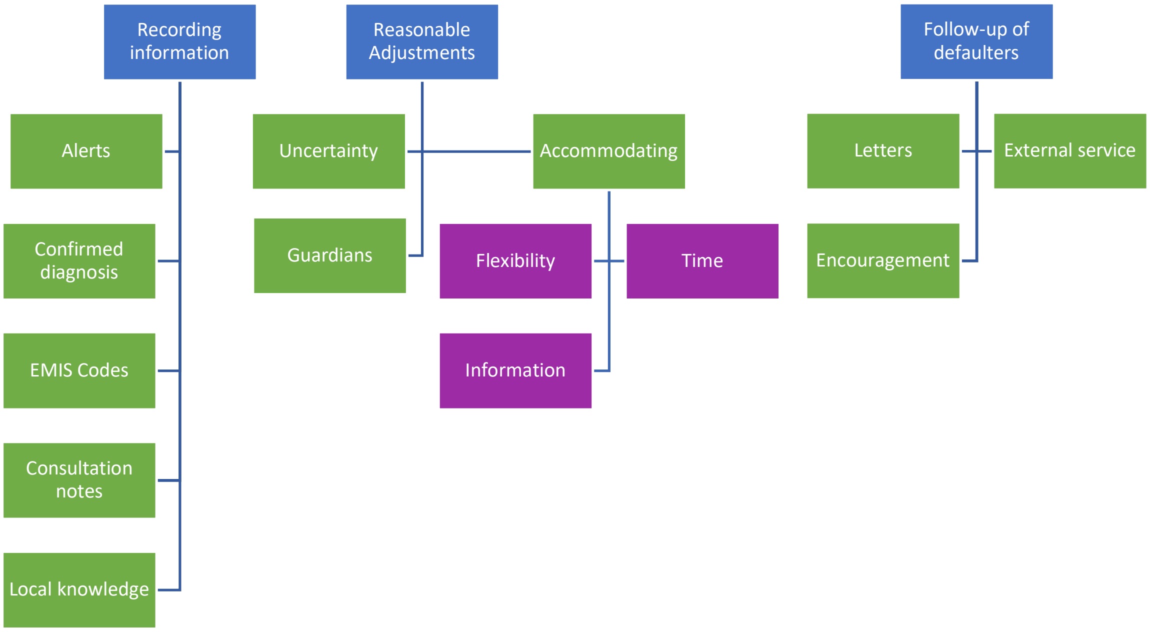
Participation Pattern	N (%)	
Single programme participation	5 (31.3%)	
Bowel only		2 (12.5%)
Breast only		1 (6.3%)
Cervical only		2 (12.5%)
Two programmes	9 (56.3%)	
Bowel & Breast		5 (31.3%)
Cervical & Breast		4 (25%)
Cervical & Bowel		0 (0%)
All three programmes	1 (6.3%)	
No participation	1 (6.3%)	

2.4 Brief Survey Information

Of the 15 GP Practices invited to respond to the survey, only six responded, all from Shetland. The responses to the questions were brief (typically one or two sentences), so analysis was limited. The coding frame from the content analysis consisted of three main categories (based on the questions asked), and 11 subcategories and three sub-subcategories were identified, across three hierarchical levels (see Figure 2). The categories and their respective definitions can be seen in appendix 4.

Figure 2

Coding Frame From Content Analysis of GP Practice Survey



2.4.1 Recording information

All six Practices reported using EMIS codes to record patients as having a learning disability or autism, but only with confirmation of a formal diagnosis. Four mentioned adding ‘alerts’ to files of patients who require reasonable adjustments. Reasonable adjustments appear not to be consistently recorded aside from any alerts placed on an individual file, and any adjustments made during appointments are recorded at the discretion of the clinician writing the consultation notes. Additional comments highlighted the small populations of people with learning disabilities and/or autism and how this means individuals are often well-known to the Practice and its staff.

2.4.2 Reasonable adjustments

Two Practices acknowledged having no procedures in place for making reasonable adjustments. One Practice reported uncertainty around procedures beyond trying their best to accommodate requirements. Responses described giving longer or multiple appointments, for example preparatory appointments prior to cervical smears, and providing more extensive explanations. Involving parents/guardians was also mentioned, as was providing written information. One Practice highlighted the benefit of being a small rural Practice and knowing the individual and their family alongside having the flexibility to alter their diary to accommodate any needs. Two Practices requested ideas for procedures that could be adopted in Primary Care to improve uptake.

2.4.3 Follow-up with defaulters/non-responders

Follow-up with non-responders varied by screening programme and is inconsistent. Non-responders to bowel screening were reportedly followed-up by letter from three Practices, although one advised this was not done regularly. Three Practices have a Practice Nurse contact patients directly regarding cervical screening defaulters, and one Practice also does this for those who do not respond to bowel screening. Breast and cervical screening non-responders are more often left to the national programmes, which are considered as external, and who deal with non-responders centrally. Four Practices described making efforts to “encourage” and educate around the benefits of participating.

2.5 Discussion

Bowel, breast and cervical screening uptake and rates of HPV immunisation in the learning disability populations of Orkney and Shetland are lower than national and local screening rates in general population. However, cancer screening uptake is comparable to other learning disability populations in the UK.

The Orkney and Shetland learning disability populations identified through Primary Care are larger than the populations known to the local authorities. This is unsurprising, given that the local authority registries are based on those using services, which are not required by everyone, e.g. care provision. The discrepancy is not insignificant, particularly with the 56 further individuals recorded as having a learning disability in Orkney and may represent individuals who require support. However, the larger figures do provide greater confidence in the data as representative of the full population in the isles.

For bowel screening, the overall rate of uptake was 57.6%, but higher in Shetland (60%) than in Orkney (44.4%). Both localities show a rate of uptake much lower than is seen in their respective general populations and also than is seen nationally, consistent with the expected health inequalities and comparative to recent data published by NHS GGC showing a rate of 44.8% and NHS Digital (England) of 50.3% in their respective learning disability populations. The higher uptake of bowel screening seen in women (e.g. Cancer Screening, 2020; Hirst et al., 2018) was not seen here, but this may be due to there being fewer older women in this population. Other patterns of screening participation were present, including delayed starts, intermittent and lapsed presentations, however numbers were small and by far the largest groups were those who never participated and those who always participated. No statistical significance was found between those who always participated and those who never participated in terms of age, gender or rurality. There are a multitude of other factors which could influence uptake however. A recent systematic review of non-participation in bowel screening identified multiple pathways to non-participation, which they organized into three main themes: differences in motivation; active aversion to screening; and contextual barriers of the healthcare system (Le Bonniec, 2023).

The uptake of breast screening was much higher in Orkney (76.2%) and comparable to national figures in the general population, as opposed to Shetland (37.5%), although this may again be influenced by the very small number of eligible women in Shetland. Still, Orkney's uptake in the learning disability population does not meet their commendable general population rates and Shetland's rate highlights a stark disparity, below the most recent uptake rates of NHS GGC's (49.9%; NHS GGC, 2022) and England's (47.2%; NHS Digital, 2022) learning disability populations. Whatever facilitators contribute to the high uptake of breast screening in the islands' general populations do not appear to apply to the learning disability and/or autism communities. Walsh et al., (2022) identified barriers to participating in breast screening specific to the learning disability population relating to a lack of understanding, difficulties with literacy and the dependency on carers to facilitate.

The population eligible for cervical screening was a more even split between Orkney and Shetland, however both island boards had very low rates of participation, with only 47.8% and 32.6% having participated respectively. Again, this is much lower than the overall national uptake in the general population in Scotland, but not as low as the uptake rates seen in the learning disability populations of NHS GGC (25.5%) and England (31%). Recorded exclusions were particularly high in Orkney (56.6%), the most commonly cited reason being ‘not clinically appropriate’, with no further detail. The temporality of these decisions is impossible to ascertain from the data available, and it is expected that individuals are not excluded without careful deliberation by the responsible healthcare professionals and discussion with the individual and their guardians. However, given the breadth of pre-established barriers to women with learning disabilities accessing cervical screening (NHS England & NHS Improvement, 2020; Watts, 2008) it may be pertinent to provide a means of evidencing what efforts have been made to overcome these, for example using standardised Read codes or screening templates, such as that used for alcohol brief interventions.

The overall uptake of HPV immunisation across the isles learning disability population was 41.9%, with Orkney’s uptake rate at 38.5% and Shetland’s at 44.4%, rates far lower than that seen nationally and locally for the general population. Previous research has shown lower coverage rates for children with learning disabilities for all immunisations, including HPV (Emerson et al., 2019). There appeared to be no gender disparity in uptake, and the zero recorded for children who had *not* received their immunisation in Orkney are likely accounted for in the 8 ‘unknown’ cases. Given that the offer of vaccination is likely to be repeated in subsequent years the uptake may still increase.

A small number of women were eligible for all three screening programmes. Encouragingly, 87.5% of women had participated in at least one programme, and a majority had participated in two. Data showed potential patterns of screening behaviour, a group of women who only participate in one programme, a group who participate in breast and bowel only and a group who participate in cervical and breast only. Data on women in the general population of Greater Glasgow and Clyde showed a lower participation in bowel screening relative to breast or cervical screening, and women who participated in cervical or breast were more likely to engage in bowel screening (McCowan et al., 2019). A recent qualitative study with women in Denmark highlighted that participating in bowel and cervical screening was more troublesome and required more steps (Kirkegaard et al., 2022). The situation may be different for women with learning disabilities, and the data may be indicative of specific barriers pertaining to either bowel or cervical screening and/or may be dependent on whether or not these women are supported by caregivers to access these programmes and the level of support available in order to overcome these steps.

Limited qualitative information was received from GP Practices on current policies and procedures pertaining to cancer screening, and there was no representation from Orkney Practices, so few conclusions can be drawn. From the responses received, recording of learning disability and/or autism is dependent on an official diagnosis, which not all individuals will have, nor indeed want. This practice may disadvantage individuals with mild learning disabilities and autistic people who have not sought a diagnosis, who may not be aware of the reasonable adjustments which could be made for them, or may not feel able to ask. There appears to be some uncertainty around reasonable adjustments, follow-ups with defaulters or non-attenders and inconsistencies in how these are recorded. Agreed processes for reasonable adjustments and how these should be recorded across Primary Care could help raise awareness of options and increase confidence in making reasonable adjustments.

2.5.1 Strengths and Limitations

While both Health Boards are comparable in the poor uptake of cervical screening and HPV immunisations, there may be scope for shared learning around how Orkney achieves greater breast screening rates, and Shetland achieves greater bowel screening rates in their respective learning disability populations. A strength of the current audit is the combining of data from both Health Boards, which has allowed a larger population to analyse patterns of uptake and given the similarities of context will hopefully reduce duplication and resource required to implement change.

There are several limitations to the current audit. Despite low levels of missing data and confidence that the dataset was likely very close to the full learning disability population, even a few missing cases could skew the results of such a small population. The low numbers and lack of data on DNA history and poor response to the qualitative element has also restricted the depth of analysis and conclusions that can be drawn.

The audit did not gather data on follow up colonoscopies, colposcopies, or other further tests, nor for subsequent diagnoses of cancer and rates of cancer found through screening, which would be of interest. Gathering the data across two Health Boards with a broad spectrum of Primary Care, and four population-level prevention programmes, was particularly challenging and dependent on the liaison and cooperation of many different teams, who naturally have differing and often competing priorities. As such, there were significant delays in extracting the data and subsequent analysis. However, the exercise has provided valuable learning for future collaborative ventures across different Health Boards which has been taken for discussion at an executive level. Finally, we were unable to distinguish between autistic individuals and those with learning disabilities in the current dataset. It is important to note that these populations may well exhibit different patterns of cancer screening uptake.

2.5.2 Implications and Conclusion

The current audit confirms previously assumed patterns of much lower uptake of cancer screening programmes and HPV immunisation in the isles for those with learning disabilities and/or autism. Orkney’s breast screening and Shetland’s bowel screening rates are outliers to the overall trend however, and further investigation may highlight areas of improvement that could be adopted by other health boards. Data on bowel screening and women eligible for all three suggests that first time participation and participation in at least one programme may be important for participating again or participating in other programmes. While gaps in the data are small, they may have a significant impact on the conclusions that can be drawn for a small population, and efforts should be made to achieve consistency in recording and follow-up, which should be achievable for smaller island boards, who also have more scope to come together and share learning.

In conclusion, this audit has highlighted that even greater disparities exist in the level of preventative care achieved for these more vulnerable island populations and should serve to intensify activities for reducing health inequalities. In order to do so, we need to better understand the experiences of people with learning disabilities and other stakeholders who may be involved in cancer screening, which will be explored in Chapter 3. Orkney and Shetland must not become complacent or relax efforts in encouraging the uptake of the cancer screening or HPV immunisation programmes because of higher rates of uptake in their general populations.

Chapter 3. A Qualitative Exploration of Stakeholders' Cancer Screening Views and Experiences

3.1 Introduction

3.1.1 Background

An ever-growing body of research is providing insight into the views of the general public on cancer screening. A recent review on public preferences around bowel screening alone included 83 papers (Tran et al., 2021). Less literature is available on the views and experiences of people with learning disability and/or autism however. A systematic review and meta-aggregation by Byrnes et al. (2020) on the attitudes and perceptions of people with learning disabilities, their family carers and paid carers towards cancer screening found only 11 papers relating to cervical and breast screening, but none on bowel screening, so findings were limited to those of women with learning disabilities (WwLD). These studies were based across the UK, but lacked specific detail on whether localities were remote or urban. Descriptions were of either broad regions which would incorporate both rural and urban localities (e.g. Northern Ireland), or counties and cities. This is an important oversight, given recent findings that throughout high-income countries worldwide, rural populations exhibit a lower uptake of all three cancer screening programmes (Walji et al., 2021). The limited data on Scottish programmes is discussed in Chapter 2.

Byrnes et al. (2020) identified ten subcategories across these studies which they compiled into four groups. The first group of findings covered data from carers supporting WwLD to attend screening and included: making decisions in the best interest of the women; creating a positive and encouraging environment for screening and prior preparation. The second group comprised of data around WwLD awareness of screening and their psychophysical experiences: women lacking understanding and awareness of screening; feelings of anxiety and fear and experiences of pain during and after screening. The third group covered professional practice barriers including the need for multidisciplinary working and an understanding of the needs of WwLD. Finally, the fourth group described approaches required to improve the uptake of cancer screening, namely: the need for education and training for everyone (WwLD, family carers, paid carers and health care staff) and learning disability-friendly health care. Byrnes and colleagues recommended future research should focus on obtaining the views of these stakeholders in order to develop a more in-depth understanding of how best to support people with learning disabilities to make informed decisions around cancer screening.

There are gaps in the literature on the views and experiences of people with learning disabilities and their carers on bowel screening, and the views of these groups who reside in more remote and rural areas of the country, who may experience different cultural and environmental influences on their decisions around cancer screening than people who live in more urban areas.

3.1.2 Current study

The current study builds on the audit described in Chapter two to provide an in-depth qualitative exploration of the views and experiences of service users and service providers from Orkney and Shetland on cancer screening for people with learning disabilities and/or autism. Four groups were targeted, using semi-structured interviews and focus groups: people with learning disability and/or autism; their family carers; paid care workers; and NHS staff involved in cancer screening. Inviting different stakeholders and combining focus groups with individual interviews allows for data source triangulation for greater breadth, participation and validity (Carter et al., 2014). The study seeks to establish a greater understanding of the local population’s knowledge and understanding of cancer screening, their experiences of being invited to participate, and how they make decisions around whether or not to participate, as well as to develop clear objectives to promote access to and uptake of cancer screening in the isles. Of particular interest is whether the views obtained are similar to those outlined in the previous review by Byrnes et al. (2020), and if contributions can be made to our understanding of people with learning disabilities and/or autism’s experiences of bowel screening. Finally, it is anticipated that the interviews will provide evidence to substantiate the conclusions drawn from the audit of Primary Care data, specifically the importance of first-time attendance on subsequent attendance, a better understanding of ‘clinically inappropriate’ cervical screening presentations and thoughts on the policies and procedures surrounding cancer screening.

3.2 Methods

3.2.1 Research Design Overview

Reflexive Thematic Analysis (reflexive TA; Braun & Clarke, 2012; 2019) was chosen as the best approach for the current study. Reflexive TA provides a means of interpreting the experiences of participants, while embracing the researcher’s position and subjectivity in relation to the project and the process of coding, analysis and theme generation (Braun & Clarke, 2022). This was felt to be important, as the current study seeks to understand the experiences of people with learning disabilities and their care providers within their unique context of living in the northern isles. The project was delivered within an NHS context using money from the Scottish Government’s inequalities fund, establishing a pro-screening position.

The contextualist stance adopted considers the ways individuals make meaning of their screening experiences and in turn how the broader social context impinges on those meanings, while retaining focus on material and other limits of 'reality' (Willig, 1999). It is important to remain mindful of the assumptions made which underpin the interpretation of the data. The premise of the current research is that people with learning disabilities or autism are less likely to attend screening than the general population, have a different experience of screening and that work needs to be done to increase participation and accessibility. The NHS is required to support the uptake of cancer screening and promote the services as worthwhile and positive. The current system surrounding cancer screening also assumes that people can access relevant information and are capable of consuming and understanding this information in order to make informed decisions about screening.

3.2.2 Researcher Description

The Primary Investigator (PI), and author, is local to the isles, believes cancer screening to be positive and worthwhile, and has a long-standing, visible position within the NHS so there was also a likelihood of being known to some of the participants by the nature of small communities. In addition, the researcher's training in Health Psychology positions her from within a behavioural change perspective. The PI was supported in some interviews/focus groups by a research assistant (RA) from NHS Orkney's Health Improvement Team. All these factors were likely to influence the recruitment and responses of the participants (see Madill et al., 2000).

3.2.3 Recruitment Process

Ethical approval was sought via the same application as described in Chapter 2 (appendix 1). The recruitment strategy took wide-ranging approaches including advertisements in local media (radio, newspapers, social media; see appendix 5 for examples); promotional discussions and visits with local service providers, including day care, respite, supported living and third sector groups; global emails through NHS communications for healthcare staff and tailored emails specific to relevant NHS and local authority staff teams. While participants were initially invited to attend a focus group with the option to arrange an individual interview should they prefer, uptake was slow, compounded by logistical challenges associated with liaising with carers and in the context of Covid-19. As such, participants were more readily recruited for individual interviews and a decision was made to promote the study based on this format in the first instance. Multiple options of contact were offered to increase participation, including face-to-face, video calls or telephone. Originally, the study planned an ambitious recruitment of 12 focus groups, with 6-8 participants in each, matching numbers across Orkney and Shetland. Due to the difficulties recruiting, this was subsequently reduced to a planned recruitment of 20 participants.

To ensure capacity to consent to the research for people with learning disability, the British Psychological Society's checklist was used (appendix 6). The checklist was initially shared with caregivers and conducted by the primary interviewer during discussions with potential participants prior to their recruitment. Participants were given information sheets to consider for at least one week before agreeing whether or not to take part. Consent was confirmed at the start of each interview or focus group. Interviews and focus groups were conducted between February and July in 2022. All participants received a £10 voucher as a token of thanks for use at a local leisure or recreational facility, which was felt to be appropriate for a Health Improvement-based project.

3.2.4 Participants

A total of 26 participants were recruited, comprising of five service users, five family carers, eight paid carers and eight NHS staff. Participants were independent of each other and were not recruited as family/dependent units or customers with their service providers. There was an equal split of 13 participants from Orkney and 13 from Shetland. Overall, four focus groups (ranging from 51 to 60 minutes) and 13 one-to-one interviews (ranging from 28 to 86 minutes), were conducted. The locality of individuals is not disclosed and names have been changed in order to preserve anonymity.

3.2.4.1 Service Users

Five participants with learning disabilities and/or autism were recruited (SU1-SU5). These service users were all female and over the age of 18 (ranging from early-30's to mid-60's). Four of these interviews were conducted in-person, with one online using Microsoft Teams. Four of the five participants had participated in at least one cancer screening programme. Talking Mats were used with only one participant, and none of the participants used the creative communication aids on offer. An overview of participants can be seen in Table 11**.**

Table 11

Characteristics of Participants: Service Users

Participant	Age bracket	Duration (mins: secs)	Mode	Living situation	Screening experience	Support Worker in Attendance	Communication aids required
SU1	40-50	36:21	MS Teams	Independent	Cervical	N/A	No
SU2	30-40	32:31	In-person	Supported Living	None	Yes	Yes (Talking Mat)
SU3	50-60	38:40	In-person	Supported Living	Bowel, Breast, Cervical	No	No
SU4	60-70	30:43	In-person	Supported Living	Bowel, Breast	Yes	No
SU5	60-70	27:37	In-person	Supported Living	Bowel, Breast, Cervical	No	No

3.2.4.2 Family Carers/Guardians

Five family members or guardians were recruited (FC1-FC5). Again, these participants were all female, four mothers, one sister and all but one was conducted via MS Teams, the remaining was over the phone. An overview can be seen in Table 12**.**

Table 12

Characteristics of Participants: Family Carers/Guardians

Participant	Relationship to service user	Duration (mins:secs)	Mode of Interview
FC1	Sister to woman in her 30s	60:10	MS Teams
FC2	Mother to woman in her 30s	72:39	MS Teams
FC3	Mother to two women in their 20s	85:56	MS Teams
FC4	Mother to two men in their 30s	35:10	Telephone
FC5	Mother to a man in his 40s	57:06	MS Teams

3.2.4.3 Support staff

Eight social care workers participated across two focus groups (see Table 13). Focus group 1 was a selection of staff from day care services, focus group 2 was from supported living and outreach.

Table 13

Characteristics of Participants: Support Staff

Focus Group	Participants	Role	Duration (mins:secs)	Mode of Interview
1	SS1-SS4	Social Care Workers	54:10	MS Teams
2	SS5-SS8	Social Care Workers	56:22	MS Teams

3.2.4.4 NHS staff

A total of eight staff across the NHS were recruited. Two learning disability nurses, two GPs and four Practice nurses. An overview can be seen in Table 14**.**

Table 14

Characteristics of Participants: Healthcare Staff

Participant	Role	Interview/ Focus Group	Duration (mins:secs)	Mode of Interview
NS1, NS2	Learning Disability Nurses	Focus Group	60:02	MS Teams
NS3	GP	Interview	35:53	MS Teams
NS4	GP	Interview	32:29	MS Teams
NS5	Nurse	Interview	43:35	MS Teams
NS6-NS8	Nurses	Focus Group	50:30	MS Teams

3.2.5 Procedure

Semi-structured interviews and focus groups were led by the PI. Questions were designed to be simple, to elicit general understanding and experiences of cancer screening, and open-ended to encourage discussion. Theory was deliberately not used in the design and development of the interview schedules in order to avoid influencing the direction of discussions. The interview schedule can be seen in appendix 7.

Interviews were primarily conducted on Microsoft Teams, which recorded both audio and video and transcribed automatically into Microsoft Word files. In-person interviews were recorded using a Dictaphone and GoPro camera, and telephone interviews using a Dictaphone. These data were transcribed verbatim into Microsoft Word, with the video recording providing additional contextual data on body language and facial expressions. A random 25% of transcripts were checked against recordings for accuracy by the RA, who also checked instances of inaudibility.

3.2.6 Materials

To account for different communication needs with people with learning disabilities, a variety of creative communicative resources were made available in face-to-face interviews, including paper, pens, modelling clay, Lego and various craft materials. In addition, the PI undertook training in using Talking Mats, a communication tool shown to support self-expression in people with learning disabilities and improve the quantity and quality of communication (Murphy & Cameron, 2008)**.**

3.2.7 Analysis

The PI conducted the analysis, beginning with an initial period of familiarisation, reading and re-reading transcripts, making notes and reflections. Coding was done on paper print-outs of the transcripts initially, before being incorporated into a specialist computer programme, Quirkos, which is designed for qualitative analysis. Quirkos allows the visual maneuverability of codes into groups which aids the generation and development of themes. Analysis is an active, iterative and interpretative process (Braun & Clarke, 2006; 2021). The process of analysis was inductive. The initial intention was to consider the development of themes within groups of participants and specific screening programmes, then generate themes across ‘service users’ (people with autism/learning disabilities and family carers/guardians) and ‘service providers’ (support workers and NHS staff). However, in practice this was felt to be an artificial distinction, which ignored the context in which people with learning disabilities and their families access and experience cancer screening: through interactions with support workers and NHS staff. Participants also spoke about cancer screening in a very general sense, unless asked about specific screening programmes. Choosing to develop broader themes across all groups and programmes generally aided the researcher's sense-making, so the analysis is presented as such. Practices such as triangulation and member checking are incongruous with reflexive TA so were not used in the data analysis (Braun & Clarke, 2023).

3.3 Results

3.3.1 Theme development

The process of analysis and theme generation is summarised in Table 15**.** Analysis was initially conducted within each of the four groups; service users; family carers/guardians; paid carers and NHS staff. For an overview of these themes and their descriptions, please see appendix 8**.** Analysis was then conducted within two overarching groups of service users (people with learning disabilities and/or autism and their family carers/guardians) and service providers (paid carers and NHS staff). Through the process of analysis, this distinction felt arbitrary and unhelpful given the information that was gleaned around the close interactions between group and the systems driving their relationships. As such, analysis was redirected across all groups, generating three main themes and five subthemes, as depicted in Figure 3. Descriptions of the final themes and sub-themes is given in Table 16.

Table 15

Overview of Within- and Across- Case Analytic Strategies

Comparison	Purpose	Strategy	Product
Within individual participant groups	Identify important aspects unique to groups	Close reading of individual interviews, reflective journaling	Coding categories, initial theme development
Within service user groups and service providers	Identify shared and unique aspects to these two groups	Data coding and theme comparison using Quirkos and diagrams, reflective journaling	Refined themes, Change of tactic
Across all groups	Compare experiences and identify configuration of themes across stakeholders	Data display using diagrams, summaries, Reflective journaling	Refined overarching themes, subthemes

Figure 3

Diagram of Themes and Sub-Themes

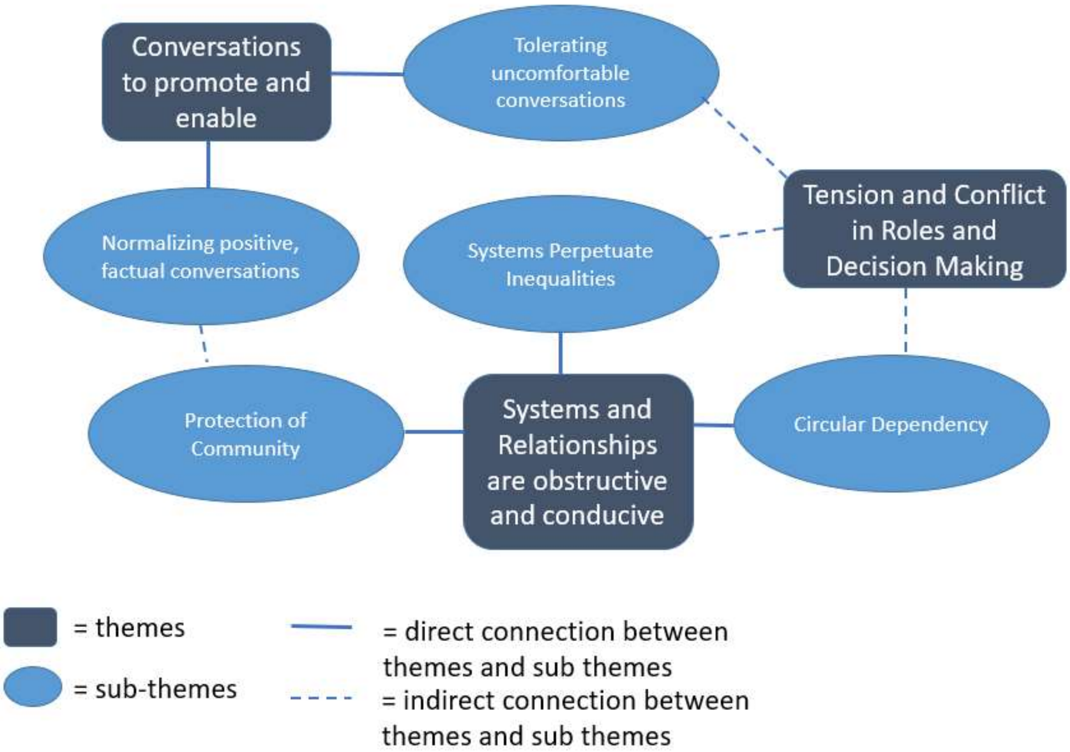


Table 16

Descriptions of Final Themes and Sub-Themes

Systems and relationships can be obstructive and conducive	Existing systems are felt to be a barrier to cancer screening, but these are underpinned by the nature and quality of the relationships between people with learning disabilities and/or autism, their guardians, service providers, the wider community and how they interact with the systems in place.
Circular dependency	People with learning disabilities/autism are dependent on their support networks for understanding and accessing screening
	Families/guardians are dependent on health and social care staff and expect them to be proactive and vigilant in their care of their dependents
	Healthcare staff and paid carers expect family carers to take ultimate responsibility
Systems perpetuate inequalities	The systems are based on assumptions that people with learning disabilities/autism are aware of the programmes, and can interact with them as they exist
	There is uncertainty about whether the programmes are open to people with learning disabilities and that the procedures are the same
Protection of communities	Small communities offer resources for promoting and normalizing cancer screening and local knowledge and awareness may act as a protective factor for those who live independently but struggle
	...But could also be a factor in perpetuating myths and stereotypes about people with learning disabilities, some of whom remain ‘hidden’, due to fears of shame/judgement from the community.
Tension in roles and decision making	The responsibility for supporting people with learning disabilities navigate complex healthcare decisions such as screening is a heavy burden and leads to tension in roles and evokes strong emotions, which influence the decisions made.
	Parents/guardians have experienced dismissive attitudes towards their instincts and feelings of protection
Conversations to promote and enable screening	Improving screening uptake involves confronting associated difficulties and discomfort and changing the discourse around cancer.
Tolerating uncomfortable conversations	Part of enabling screening is about becoming comfortable with uncomfortable conversations and implications; being open and honest about the unpleasant and more difficult aspects of screening
Normalizing positive, factual conversations	But also balancing this with early and ongoing education and positive conversations about screening as well as tackling the stigma and myths which still exist about people with learning disabilities and/or autism

3.3.1.1 Theme: Systems and relationships can be obstructive and conducive to cancer screening

Systems perpetuate inequalities. Cancer screening is dependent on various health and social care systems working and interacting effectively. However, the processes of cancer screening, and the wider systems they feed into, may perpetuate the inequalities faced by people with learning disabilities or autism.

For cancer screening to be effective, the system assumes in the first instance that people with learning disabilities or autism, and/or their carers, are aware of screening. The participants in the current study who have learning disabilities or autism all had some awareness or experience of cancer screening and are likely to be a poor representation of the general learning disability/autism population in this regard. Only one autistic woman described independently interacting with screening. Both support staff and family carers expressed misgivings about people with learning disabilities' knowledge and understanding:

> *PI: OK. Well, what about with the people you support. What do you think they know about cancer screening programs? [Respondents all shake heads]*
>
> *SS7: Nothing*
>
> *SS8: Probably very little*
>
> *SS5: Yeah.*

There was uncertainty as to whether the invitation process is the same for people with learning disabilities or autism, even within healthcare staff.

> *I'm curious as to how… people with learning disabilities are invited, but then, like how you said they're maybe not coded as learning disability. - NS7*

-

They'll get, I would imagine they're gonna get the letter the same as everybody else?...I don't know if there's access to em, if they have access to anybody you know, is that something like the learning disability nurse or anything would help with? I don't know... - SS2

Lack of awareness of eligibility for cancer screening also existed within family carers:

I didn't know those with learning disabilities would be invited. I thought it was just normal people. That sounds awful. But, you know. – FC5

The above quotes reflect that people with learning disabilities and autism are still perceived as 'other'. Different services and pathways are experienced and expected as a standard, and for this reason some families caring for people with learning disabilities or autism and those with learning disabilities or autism themselves may dismiss cancer screening as not relevant to them. This may explain some of the difficulties in recruitment for the project, but also highlights the potential benefits of tailored advertising for specific populations.

Cervical screening was a particularly divisive issue in this regard with the majority of participants citing a lack of sexual activity as a reason for not going ahead with screening, and a few participants arguing there are still too many assumptions made about people with learning disabilities or autism and sex, and that for non-autistic or learning-disabled individuals, sexual activity would not be in question.

Despite these uncertainties, people with learning disabilities are expected to follow the same procedures as the general population. Implicit in these quotes is that this is challenging without adjustments. The learning disability nurses were explicit in their view:

I think the the way they the, the, the screening projects are set up or the way, the screening, access to screening is set up almost isnae... I- I suppose is a health inequality in itself because, em, how… there's just that- that they follow, people with learning disabilities expected to follow the same kind of procedures as the general *population in relation tae, and what I mean by that is fit intae a 20 minute appointment to get a, a cervical screen, ehm, and eh, to the bowel, the bowel screens are sent to their houses and their expected to be able to read, interact wi' it, complete it and send it off. Now, that's four different tasks in one. I think it needs to be there has to be a separate procedure for the general population. I think it needs to be moved away and foc- and there has to be a focus on people, people wi' learning disabilities.- NS1*

Invitations rely on the receipt of, and engagement with, letters which are sent to eligible individuals, but there are significant problems here before screening can occur:

I just dunna budder opening my mail – SU2

-

An' they might be completely unaware that they've actually received this letter {SS2: Yeah}… I guess initially [supported living staff] would speak to the family, the guardian, to say they've received this letter as opposed to just opening the letter an' mibbe immediately talkin' it over wi' the eh, service user you see, so. Maybe even, I don't know, but mibbe even it's getting shut doon at that point you know? – SS4

-

…this whole thing of things being sent to them in the post, that they can't deal with. {PI: Yeah} And, you know, there's no system for that, it's just like people have to get them off their desks, and you know, pass it out, there's no follow up with how it go- so this would be an area where, it would have to be a clear situation. – FC4

-

Recipients in charge of their own mail need to be motivated to open and understand it. Where family or paid carers oversee an individual's mail, they too also need to be able to understand, but also need to be aware of screening's relevancy and feel it is important and feel able to discuss this issue with the individual and others who may need to be involved (see section 3.3.1.4 Normalizing positive, factual conversations).

Non-attenders may be picked up in some proactive GP Practices, and while some procedures are in place to offer reasonable adjustments, such as registers of people who have learning disabilities or autism, phone calls or extra letters to encourage individuals to come in to discuss; there exists considerable Practice variability and information gaps.

> *Invitations, calls and recalls, all of that stuff is taken care of from outside the Practice...In a previous Practice I worked in... If people had not participated in the screening program, we would contact the patient and say. We noticed you haven't been. Do you want to talk about it? Is- you know, what are your concerns or expectations, or is it, you know what, what to do? And if the patient chooses not to attend, that's absolutely fine. But I think the feeling was. That people might be more likely to respond to their own doctor. {PI: Yeah} So, so, but we don't do that here. Umm, we don't. We don't do anything really here. No. So we just rely on it to run in the background. - NS3*

The above quote highlights an awareness of the importance of the GP relationship but also the general detachment of screening from Primary Care and an expectation that the national programmes' communications are sufficient.

There is an awareness of the barriers which exist for people with learning disabilities and the need for more intensive engagement, but the systems feel rigid and options are limited. Information about the individuals, time to check their notes and time within appointments to explore issues with the individuals are cited as critical but limiting factors.

> *I don't consistently go and look at each person on our e-mail [sic] system to see if they have autism because I don't have the time for that... See when when they come to me for other things that I don't have to time, I have got a very short appointment time for, for that element of care I'm doing at that point. So I don't have that time to look into it even deeper and think ohh perhaps we need to have a discussion about this em, also sometimes I don't know until they come through the door that they've got a learning disability 'cause it's not coded. – NS5*

-

I mean, I don't, I don't always think the information is that accessible if you don't have a learning disability. So you know what it means. Screening is a complicated thing to explain, especially in a leaflet… - NS3

The reality is that awareness of screening for and in the learning disability and autism populations cannot be assumed, and invitation letters are likely insufficient to encourage or prompt engagement.

Circular dependency. Participants with learning disabilities are largely dependent on their support networks, in particular their carers and GPs, for their knowledge and understanding of, and access to screening.

I never go to the doctors by myself 'cause the carers help me. – SU4

-

Ach, I think it was one o' me carers that telt me aboot it [breast screening]? – SU5

-

PI: Whit do you think wid mak it easier for folk to go an' do [cancer screening], whit wid encourage dem ta go?

SU3: Tell dem? {Ok?} An spaek aboot it, you go ta doctor go an' spaek aboot it... I think go an' see da doctor {yeah} aboot it.

Family guardians are also dependent on professionals, and express expectations of proactive action from health and social care:

I think there's just laek level of. Trust or somethin' that. Somebody is going ta send you a letter ta say that she needs something, in terms of her meedical needs. Em. 'cause, we're not. A meedical family… we would be reliant on da doctor ta be sending, so if Gemma was due something we would be trustin' that the surgery, well the health centre, sends oot a letter ta say that she was due somethin', basically. – FC1

-

… When we’re away, somebody would have to take on all these things, as well as the cutting of toenails, and yeah.... I think the GP or the nurse would need to bring it [screening] up with them at the next, eh, visit. - FC4

These expectations are all-encompassing, including a vigilance and level of detail so foundational as cutting toenails. Many families of people with learning disability or autism are after all, used to an invasive-level of multi-disciplinary professional involvement and observation, described by one mother as like being in a ‘goldfish bowl’ (FC3). Paid carers feel the pressure of expectation via family or guardians’, especially as care defaults to them in the absence of family guardians, and express worry about their influence on decisions around screening.

Kind of worrying for us as well em. [the family are] very “ah well, you'll know if there's anything wrong because you know that person really well”. So it kinda puts a good bit o’ pressure on us too? And because you're proactively looking for problems [laughs] when there's possibly none em. But yeah, so that that's their kinda logic. That's like. Well, it'll be OK because you'll know if there's anything wrong. – SS7

-

I certainly think they [supported living staff] can influence an’ sometimes family members look for that because they don’t know, they don’t know- or they don’t feel like they know the right answers or the right decisions {SS4: Yes} so sometimes they actually look for sort of advice or “oh what do you think” and, you know, and they can be influenced by, by the staff. – SS2

These dynamics – the question of responsibility, coupled with an absence of collaboration and ‘right’ answers, or the potential for irresponsible influence - promote an environment of hypervigilance and doubt around the healthcare of people with learning disabilities.

As the quote from FC1 highlights, there are also significant expectations placed on Healthcare staff, in particular GPs – who have relatively little to do with screening programmes – to discuss these issues and take ultimate responsibility. Healthcare, of any kind, is synonymous with doctors. However, healthcare staff expect families to share information and take responsibility for themselves.

…ultimately they [family carers] are the only people that can make the decision. We are here to help them, you know, shared decision making is what we should be doing and we are here to help provide as much information as is appropriate or helpful but- but we, we we can't, it would be assaulting [laughs] that person if we we just decided to immunize [for example] and and there was no consent for that.- NS3

-

…we need to get to know the person and often as well we are relying on family and support workers for that information because it often at times you don't have that time tae build up that therapeutic relationship... It's no a short term piece of work. Please, please don't get me wrang wi' that. It's definitely something that can take a long period of time. So you're not talking about a 20 minute appointment. - NS1

The question of where responsibility lies remains up for debate, with parents and carers questioning the issue themselves.

Noo, when that bairn goes into hospital, then the parents are quite often best placed to deal with that things an ony specialist care that bairn has because they're dealing wi' it on a daily basis… So some of the care will be devolved to the parent. But ultimately, the responsibility for seeing that care has taken place goes back to the medical staff. And it's like that,[weighing up with hands] that kinda, whaur does that balance sit because it's the same kind of thing. Whaur does the balance sit? – FC3

-

That's the thing. Who's responsibility is it? Is it everybody's conversation or is it purely for… You know, respecting privacy of other people, to them it's their decision, is there anonymity there? I- this is what I don't know, especially in- in, you know, I don't have much experience in this area, but I just wondered was what [SS8] said. You know. Is there may be a gap there in that conversation? – SS5

The latter quote polarizes decision-making around cancer screening to engage in a joint conversation or respect it as a private decision and highlights an uncertainty of how to unite these perspectives. Another view is that Health and Social Care staff *do* need to step up and take some responsibility, particularly in cases where there is little or no family/guardian involvement. One way this is being delivered is through the establishment of annual health checks for people with learning disabilities.

I think it's difficult and I think that's part of the reason, ri- really why we've been trying to start doing this yearly review because I think you know no one, it is like you say, hard to know who- who's responsibility it is for, you know, bringing them in for a problem or trying to pick up problems and things like that. I think yeah, because it's hard to know who should be doing what. It's just easier to bring them in one time and try and cover everything as well. I think, you know, if they are then used to coming yearly to see the nurse and then the GP, they build up that kind of relationship as well. So it's not such a big scary thing to do. – NS4

-

Sometimes we need tae intervene, and it's our role as nurses and it's support staff or families… responsibility to make sure that that person's got the best information available… It's our responsibility, as caregivers, to give them the best advice possible. So this… myth that we should allow that person to make decisions unilaterally, without any kind of, em, responsibility is is a disgrace to even think that way, because we would never do that for somebody that we loved or cared for. – NS2

The emphasis here lies in proactive, engaging care, which sits in contrast to the passive expectancy of public screening interventions. Of particular consideration is bowel screening, which is conducted solely at home; uptake may benefit from more proactive involvement from Primary Care.

The components of the support network operate independently and exist in something of a circular dependency: each are dependent and expectant on another to some degree for direction and decisions. What is evident across interviews is a solution-focused attitude, and a desire to work better together. Perhaps the argument of *who* takes ultimate responsibility is distracting from the real issue of how to engage everyone taking a more proactive, collaborative role towards enabling cancer screening.

Protection of community. Many participants in the current study described communities of trusted, quality relationships including third sector services, activity clubs, employment, peers and neighbours. Interactions were praised for being nurturing, proactive and strength-based, focusing on building confidence and motivation. Communities can act as a protective factor around individuals with learning disabilities or autism, relieving the burden on family carers, compensating for gaps to help identify those in need who might otherwise be missed: individuals who are not in support services or who do not have a formal diagnosis of autism or learning disability.

> *There's another group of folk who, of people with learning disabilities who actually live independently, and mibbe with minimal support from either family or, you know, sorta paid staff. An' I'm just thinking if, they- they'll receive the letters too and might not even understand what's in- you know, might not be able to read the letter properly. – SS1*
>
> -
>
> *...one of the advantages of being a small community is that we do know our patient population pretty well. So those people who do maybe need just a bit of an extra support or like telephone reminders about their appointments and things like that are usually picked up. - NS4*

Communities may present a valuable and under-utilized resource to promote and normalize conversations around cancer screening. A learning disability nurse described an intervention they delivered in a previous role, designed specifically for one individual living in supported shared accommodation who was at increased risk of testicular cancer:

> *It was "Test Yourself Tuesday" they called it, which I thought was brilliant...they ended up doing it with the whole house. Every man that was in the place, and they would all come down and go "No never felt anything!" And you know... and it became this…thing that they've done...it was somebody else in the house who through this process said "I*

think I found something" ...And it turned oot he did have testicular cancer and he found it so early. - NS2

The sense of community, fun and openness feels core to the success of this intervention. A contrasting view of small communities is the idea that some people are more private and withdraw more because they are more visible:

… it's a small place, so some families do do talk and some families don't want to talk... - SS6

-

I mean we work in remote and rural areas an' my experience o workin in some remote and rural areas [laughing] is the people wi' learning disabilities, they're no exposed, they're kept in the house, you know, they're kept, they're hidden away [gestures "over there" with hands]. - NS1

There may be a fine line between protection and concealment. Interactions with healthcare systems, the wider community and local cultures may feed into other issues, such as perception of healthcare for people with learning disabilities and autism, as well as myths and stigma around these populations.

3.3.1.2 Theme: Tension and conflict in roles and decision making

The family carers of people who require support describe living within a ubiquitous system, surrounded and managed by multiple professionals. These interactions can feel bureaucratic and critical, and set a script and a tone for what families expect when navigating wider healthcare systems.

...You still hae to be the one that shouts. {Yeah}…It's a bit laek being in a goldfish bowl sometimes because you're aware that aa this folk ir aa looking into your family...And-a. So you kinda think. Am I doing the things that they'll think is right? Am I doing enough?... Kaen, it's it's a. I don't know. It's funny. It's funny what you hae ta... How-how it aa works. - FC3

This family carer also laughingly recounted a story about a professionals meeting she attended with her daughter. She described a 'proliferation' of such meetings during this time which felt ‘tokenistic’, where reports were not shared with her in advance, and she was forced to think on her feet. At one of these meetings, her daughter ran over to a window, opened it and proceeded to shout “Help!” to passers-by, which her mother thought was very funny and felt indicative of their feelings of frustration and the almost farcical nature of the meetings. Despite being closely observed, people with learning disabilities and their family carers still have to shout to be heard. A core assumption of screening is that individuals make informed decisions and actively engage with the process, either independently or with support. In contrast, families and people with learning disabilities can feel disempowered: their general experience of health and social care services is that they take the lead.

The burden of responsibility for decision-making around cancer screening stretches the boundaries of individual roles, evokes powerful emotions and involves significant challenges in navigating consent. For family carers, considerations around cancer screening for their dependents are seen as another aspect of a parenting role, but the complexity of such decisions amplifies the tension between the fierce parental desire to protect and save from further suffering, and a diminished ‘guardian’ role which allows their adult children independence and right to choice.

....It's hard as a parent when your child is an adult and at the moment you know everything seems to be Alice’s choice and um, and that's hard when you're trying to steer her towards making healthy choices…my problem is I can't let go because I have to, have to trust the carers that are looking after her to be having the eyes of a mother. And nobody does have the eyes of a mother. - FC2

Family participants are aware of their own dependency on carers and healthcare staff. The above quote is illustrative of the conflict provoked by that dependency: it describes the experience of trusting others as an obligation, illustrating discomfort in relinquishing responsibility to paid carers and a feeling that it is not enough.

Parent participants described how protectiveness and vigilance towards their son or daughter’s health has been viewed negatively by support staff and healthcare workers. In some instances, it has been used against them and even as a cause for dismissal despite valid cause for concern.

> *…It's not helpful for the parent because sometimes, you worry because you know that something's not right, and they're [doctors] not listening. And they make you feel like you're overreacting, and then they find out you're not overreacting, so- overreacting, you're not an overprotective mum. – FC5*

The conflict is exhausting. Both healthcare staff and support workers hold compassion for the families they support, recognising their burden and where this causes tension in their own roles. Challenging others feels like an ethical dilemma between a duty of care to the people they support, and respecting the decisions and privacy of the family and guardians in order to maintain trusting and positive relationships.

> *[It makes me feel] helpless sometimes. Be- you know, because obviously respecting people's wishes, but at the same time. I- I don't know, but in my I can only speak for myself. You feel like, are you giving the best care? - SS5*

Decisions are often felt to be hasty, made without even attempting screening, with expectations that the individual won't cope with the test or the potential negative outcomes.

> *You know that they there's just always this assumption that, oh they wouldn't handle going for smear appointment, or a breast exam or or anything like that and it's just automatically no they're not going, whereas, you know perhaps that person might actually, em, take it in their stride, and sometimes there are a lot more resilient, our service users as well, {RA: Mmm} but they perhaps cope with going to that appointment better than, you know, some- some of us, you know what I mean? - SS8*

These decisions may be driven by uncomfortable projected feelings of those facilitating screening; There is a sense of fear, primarily for getting things wrong.

I think it’s partly because they- it would maybe be their [carer’s] job to support the person to go to their appointments and they’re maybe thinkin’ aboot their own feelings around that? As opposed to their own views about how important a screening is. - SS1

-

You know, what I really feel very uncomfortable about. It [cervical screening] felt very uncomfortable. And, you know, as a- as a- as a- as a nurse myself, I- I really felt I was. I was on, you know. I- I was on thin ice. I feel, you know, I'm. I'm I really. I- I didn't. I didn't feel comfortable having to make that call my-mys- I'm to say. No, I am. I am not doing anymore. – NS8

In the case of cervical screening, which is generally overseen by Practice nurses, there is some debate as to whether responsibility should be laid on GPs with expectations that the screening is not likely to be appropriate, or possible.

…I think with- with someone with, with someone like that, that it's- it's almost more appropriate that they come and see a GP before they come and see a nurse if poss- if we already know, you know, that they're on a- a learning disability or or- or- or- or the- the some autistic or that, so that the- the doctor can- can save them from any. Em you know it. It's almost like a- a. Do you know what you- you relying on- on their- their carer or their parent and yourself judging when to stop, you know and- and when to stop trying and- and eh. Yeah, I I just hope, you know, I just get that I sometimes wonder, should we have been even having it in the room on that on that table, you know, and- and- and I… You know, I- I thought it with him. I- I felt that a-a good in consultation with a doctor would- would have em. You know the- the appointment with the just simply wouldn't have happened… - NS8

The above quote from a Practice nurse argues for due process involving the GP in cervical screening from the outset. There is clear sense of discomfort and fear around breaching a boundary, and a feeling of ‘doing something *to* somebody’. Despite two lengthy preparation appointments, expressed consent and the presence of the person’s mother, this example ended in a consultation with a GP, who ‘saved’ the person concerned by concluding that cervical screening was not required as there was no sexual element to the woman’s relationship. Which begs the questions, is a lack of time and support the primary issue, as touched upon in section 3.3.1.2, or rather healthcare workers’ fears, embedded assumptions and stigma around healthcare for people with learning disabilities and the hierarchical systems we work within? NS6 challenged her colleague’s view:

…Equally I kind of think that if... Like as a- as a person. If you've got a learning disability or you've got autism, that regardless of what the- what anybody else feels, if you want to actually hiv that screening, we should, we should try and enable you to have it. – NS6

This theme feeds into the broader discussion of systems and relationships, contrasting the view of an ultimate decision-maker with the ideal of everyone supporting people with learning disabilities and autism. True shared decision-making allows a careful balancing of the burden of choice and risk, empowerment and duty of care. There is a real risk that informed consent and shared decision-making become lost in the overwhelming, conflicting feelings of those whose primary responsibilities for the adults they support should be educating and facilitating screening.

3.3.1.3 Theme: Conversations to promote and enable screening

Tolerating uncomfortable conversations. The entire process of cancer screening, from navigating the system, experiencing the test and waiting for results, requires tolerating a spectrum of distress, ranging from mild nuisance and discomfort through to real fear and significant pain. At an individual level, in-depth preparation is required, and information needs to be tailored. Conversations need to be gentle and reassuring in order to normalize the topic and increase confidence. Those familiar with individuals are best placed for these conversations, as time, repetition and quality relationships are essential conditions. A multi-disciplinary approach is also critical.

Several accounts described expressions of fear or worry around the merest mention of cancer, with a strong association with death, but also anxiety around medical interventions more generally. It is worth noting that the population for which this is true would be very unlikely to have participated in the project, and their views are therefore missing.

I think for somebody laek Gemma, ony-ony mention of cancer laek even an advert on the TV that's raising money for Cancer Research or something, she'll go, "oh, don't say that word!". She's got a really, laek, she doesna even want ta hear it. - FC1

-

It's how you support them because their understanding, their feedback on pain and discomfort. Em, and the general knowledge is different and difficult for them sometimes to understand without them getting totally paranoid, am I going to die? - FC2

-

...laek we geed for the vaccinations, what she thought she was going- as soon as she saw the injection, her first thought was they were going to try and tak blood fae her. And we haed ta spend, kaen, oh at least 10 minutes goin, an'- there was, there was- [as if speaking reassuringly to Martha] nae blood, there was nobody was going tae tak ony blood fae Martha at all. - FC3

At the upper limit of distress our human threat-response comes into play which was evident in some of the interviews with people with learning disabilities, particularly around cervical screening. Some dissonance exists here, as participants can objectively recognize difficult feelings and maintain their stance on the importance of screening and other health interventions, but protect themselves by closing, avoiding or distracting from discussions.

SU3: I think I, eh, I – I think I had dat [cervical screening] {Ok} a braw while ago {OK} but am no haed it again.

PI: Ok so you've just haed it da wance?

SU3: Haed it da wance.

PI: Yeah. Can you mind whit dat wiz laek?

SU3: Eh…Strange [short nervous laugh]...An' it's odd. [continues flicking through leaflet]. Very strange. {Mmhmm}. But it's aaright.

PI: Can you mind anything else aboot da experience? Aboot going?

SU3: Mmm.[speaking very quietly] Just goin' der an' comin' back an'. It joost [pause 3 secs] I don't know. [sighs, indicating a wish to stop line of questioning].

This same dissonance may also be an issue with health and social care workers who are required to facilitate screening, as some of the above quotes have already touched on. In addition, some family carers of people with learning disabilities and autism also need support to consider sensitive issues such as cancer screening.

Some families are are not strong, they're not strong enough to cope with that themselves. And then having their child go through that too. And I think that's where they have that control still. And it's about those things that they can control. And not for the right reasons sometimes. - SS7

-

So when [SS7] was saying about families being private, that could be seen, you know, that's not something that's mainstream, it can be seen as being something <u>too</u> big, too fearful to face whatever that step is. - SS5

The fear of harm, of unwanted outcomes and of going through the process of something inherently uncomfortable is not limited to the person invited to participate in screening, and neither are the conversations. Gradually broaching these difficult conversations in safe contexts and within safe relationships feels essential if the goal is to instill a sense of control for all involved and a sense that screening can be a part of normal healthcare. For example, participants often related screening to their own or others' health problems or healthcare experiences they've had to endure, and when asked about methods or ideas for coping, most mentioned talking things through with trusted others.

I spoke to me friends {ok}, they reassured me that everything was alright. - SU5

While other reported methods of coping with screening were more adaptive (e.g. humour, seeking reassurance, relaxation) than others (e.g. avoidance, general anaesthetic), this line of questioning felt more conducive to positive conversations about screening with *all* participants, allowing a more solution-focused, personalised way of thinking and providing foundations for a sense of control rather than a focus on discomfort.

Normalizing positive, factual conversations. Ultimately, promoting screening begins at a population level. Cancer is highly visible in small communities, bringing with it strong, negative connotations of fear, illness and death.

But when you come heem, somebody's got it or somebody's recoverin' fae it or somebody's deein fae it, and it just seems ta be everywhere, here. And it, that mibbe is annider community type thing 'cause you keen everybody is linked. And that's really interesting. It's interesting for somebody laek Gemma it's obviously lost quite a few family members have been really special til her. But she also she's probably al-al laek, always aware o somebody wi' it an' it's no a good thing. - FC1

-

Well I know it's not nice to get it, I do know that. {Yeah} Mm, people can die with it. Even, even wee children with it so and they're only young bairns {Yeah, yeah} see on the TV all the time, peerie bairns get cancer. It's horrible.- SU5

Discussions are illustrated by fatal cancer experiences of friends or family members and members of the community, while stories of recovery are largely absent.

I had a friend, [name], now she used to go tae [day care] with us, she was in our class. Well. She got eh, cancer, now it was stomach cancer she got I think, and the nurses and doctors and surgeons couldn't help her at the hospital, so they pat her to the [care home] and she d- only was there a few days and she died, just like that. - SU5

-

My stepmum died of cancer about a year and a half ago…. It... If you don't get it in time, it can be really quite horrific. - SU1

There is a strong awareness within discussions with health and social care workers of the need to impart knowledge and education and engage in positive conversations around screening. Information needs to be in the public domain, presented earlier, in multiple formats to raise awareness and normalize screening in order to instigate open conversations across society.

Even if you could get together like a. Like a family group session and, you know, verbally give them the information, the families and it might put them at ease a little bit instead of giving it in black and white letter… cause that can be quite hard to take on. And I think sometimes speaking in a group session when they're not isolating thing on their own, reading that letter. Em then you can all chat about that. - SS7

-

I think that there's a massive training elements tae it, a massive knowledge element tae it. There's also a bit how we communicate, why this person needs to have a screen, you know what are, a detailed approach to what the screen involves. Em, do we need to be linking and we public health. Do you know, an' a- I think and also like [NS2]'s example, how do we, how do we make it trendy? How do we make it kinda… You know, em fun, easy. Uh, how do you make it accessible? - NS1

Seeing information as mainstream can tackle fear. When it comes down to going ahead with screening, the majority of participants see it as positive and essential, highlighting the speed, ease and resulting relief and reassurance from a negative result.

It's good ta hae yun, an it's good ta go tae yun, an' it maks you kinda, sa- it maks you, you safe, it maks you keen kinda ten times better yourself. More happier.- SU3

-

I'd say it's better for you to get it done and send it off, so you can know you're clear. - SU4

-

It seemed to be clear so {Ok} thank god, that was a mercy! {Yeah, yeah} I didna hae onything ta worry aboot! {Yeah} 'Cause if they telt me it wisna clear, well I wida been even more worried - SU5

-

For me, it would be important that he had it done. And. It just gives you peace of mind. - FC5

However, there exists an artificial black and white distinction, which leads to screening being seen as something of a litmus test for cancer and impending death. This in turn can lead to the fear and avoidance described above.

Do this quick and easy thing. To... prevent your chances of dying of cancer, I mean, you know, that's it. - SU1

-

... I would go ahead wi' it rather than get cancer, I think... I know screening is best for you. And you know whether you've got it or not. That, that's the thing you see. [big sigh] - SU5

Experiences of stigma may play a role here also, unconsciously ingrained in views of people with learning disabilities and their families, and how they will be treated in healthcare settings:

Well. [pause 4 secs] So one of the doctors, one of the consultants, told me that if [son] had a tumour, they wouldn't treat him. And, another one said, "Well, you wouldn't want to put him through that, would you?" Em. "You don't get any pain". – FC5

Learning disability nurses who are working across community and health settings are concerned about the level of ignorance and pervasive, derogatory myths that still exist, describing a variety of examples which make the above quote less surprising.

Ultimately, we still have a long way to go in changing the culture of how we treat and think about disabled people, particularly in remote and rural areas where the populations are described as being kept “hidden away”.

3.4 Discussion

This reflexive thematic analysis provides a synthesized overview of the broad issues pertaining to the understanding of cancer screening programme uptake in people with learning disabilities and autism in Orkney and Shetland. As the analysis was conducted by a Trainee Health Psychologist working within an NHS Public Health setting, it is important to reiterate the pro-screening stance taken from the outset. All data have been interpreted with the view that screening is positive and people should be able to find out if they have cancer as early as possible to prolong quality of life and reduce mortality.

The assumption that population screening programmes are sufficiently accessible to all is problematic when applied to the learning disability and autistic populations. There is a dependence on others to facilitate education and access to overcome systemic barriers to screening, but uncertainty remains around invitation processes and whether screening is even applicable to people with learning disabilities and/or autism. The result is a circular dependency and a lack of leadership. A recent systematic review found multiple systemic barriers to informed consent in healthcare, including time constraints, inflexible models of care and insufficient support for people with learning disabilities and health professionals (Dunn et al., 2023). Power et al. (2022) argue that the current social care agenda of personalisation – aimed at increasing individual choice and control – ignores the relational nature of care and support and increases inequality by placing the burden of responsibility on the individual and their family. This context suggests a paradox which contradicts modern conceptualisations of disability, such as the social and human rights models of disability, both of which locate the “problem” of disability as existing outside the individual and within the environment or society, rather than a problem with the individual (Lawson & Beckett, 2020). Cancer screening programmes need to take reasonable adjustments much further than easy-read leaflets to acknowledge the relational nature of healthcare for people with learning disabilities and autism. This may require more proactive input from health and social care support networks. Changing the systems for people with learning disabilities and/or autism, for example through targeted community cancer screening events, may be more successful than increasing efforts to engage these populations in the current systems.

The close-knit communities of Orkney and Shetland may support cancer screening through their local knowledge of potentially isolated individuals. There are also benefits from smaller GP Practices who more often have time and resources to follow-up on missed screenings. However, these practices are inconsistently applied and living in smaller communities significantly increases the visibility of difference, leading some families and individuals to keep to themselves and reject offers of support. There is a lack of current research on the impact of smaller communities and geographic factors on the lives of people with learning disabilities and/or autism and their health outcomes. Insufficient attention is given to geographical factors in health research generally (Wark, 2021), and particularly so within intellectual disability research (Wark, 2020). A 2013 study examining the impact of remote or urban living on social exclusion in adults with learning disabilities in Scotland and found that those living in remote areas had better opportunities and were less deprived than their urban counterparts, but contrary to expectations they were found to have poorer quality relationships, possibly due to their social network comprising more of paid staff (Nicholson & Cooper, 2013). Community, in the current study, was felt to be an important resource for people with learning disabilities, and provides potentially valuable opportunities for opening and normalizing conversations around cancer screening.

Decisions around cancer screening are complex and difficult, and discussions highlighted tensions within relationships and individual roles. Parents in particular struggle with the conflicting messages they receive about what their role should be with regards to their dependents' health, the emotional burden of causing distress or missing potentially lifesaving interventions and experiences of judgement from others. Practical tools and guidance for family members and health and social care workers in supporting decision making may relieve some of the burden of responsibility, aid the juggle of rights, risks and practicalities (Bigby et al., 2019) and promote genuine informed consent. New developments such as the La Trobe Decision-Making Practice Framework (Douglas & Bigby, 2020) show promising outcomes for both people with learning disabilities and those supporting them in making decisions (Bigby et al., 2021).

Ultimately, there is a need for a shift in conversations around screening, which involves bringing them into public discourse earlier, in familiar and encouraging contexts and safe relationships. Misinformation and discomfort around having open conversations around cancer screening can lead to anxiety, fear and ultimately avoidance of screening, even where individuals feel it is a good thing to do. Fear around cancer, the prominence of narratives around negative cancer outcomes and screening avoidance have been explored previously in the general population (Vrinten et al., 2017). Metaphors such as the 'war on cancer' are familiar in mainstream media and may obstruct population-based prevention efforts, through increasing worry and decreasing the sense of individual control. Education around cancer and cancer screening needs to be tailored to the unique needs of people with learning disabilities and autism. Education which begins early, provides a balanced approach to both positive and negative aspects of screening, is fun and engaging, and is conducted in familiar and non-healthcare settings in the company of peers, may aid normalization and reduce misinformation and fear. Education and training for health and social care staff is consistently recommended in the literature on healthcare for people with learning disabilities and/or autism (e.g. Andiwiyaja, 2022; Byrnes et al., 2020; Chan et al., 2022) but may require more committed action from policy makers to ensure implementation.

The current study broadly supports the findings extracted from Byrne's (2020) systematic review, which highlighted fear, worries over pain and the influence of family and paid carers as barriers to participating in screening. Education and implementing reasonable adjustments were the two main factors identified to improve uptake. The current study serves to reinforce these recommendations, but perhaps takes things a step further. This study highlights the relevance of education for other stakeholders, such as social workers and third sector or community-based services, and identifies specific issues which should be incorporated in any training or educational sessions.

Particular attention should be given to pervasive myths around people with learning disabilities and autism, such as whether screening is applicable to this population, especially with cervical screening and beliefs around sexual activity.

Multi-disciplinary working was identified as an important facet of ensuring the health of women with learning disabilities, but specific interventions and guidance may be required in order to aid and facilitate Multi-Disciplinary Team (MDT) approaches. Encouraging open conversations about screening may need to be preceded by support for families in how to navigate complex decisions.

3.4.1 Strengths and Limitations

The strengths of the current study include being the first study exploring cancer screening uptake in the learning disability and autism populations of remote and rural Scotland. The study provides a broad overview of the many considerations of different stakeholders involved. While participants were largely positive about screening, support workers and family carers were able to give some insight into the population who are not pro-screening and would have therefore been difficult to recruit.

There are several limitations to the current study. Only people who were open to engaging in screening participated; so the views of individuals who choose not to access cancer screening are missing. Participants may not be representative, for example, the majority of service users in the current study live in very similar contexts (supported living accommodation), and only one lives independently without support from paid carers. This individual presented a very different experience from the others, however as this was only one view it is impossible to establish any pattern in interpretations. Another significant caveat is all participants were female, except for one staff member. While a female majority was expected, given the target populations of the screening programmes, men may have significantly different experiences. It was also not possible to include a discussion around HPV immunisation due to the ages of the participants. Finally, the views of social workers are felt to be an important oversight, given the integral role they have played in many examples given.

3.4.2 Future research

Research on the views of those missing from current study will be important, particularly men, and those who actively choose not to participate in screening. More attention should be given to geographical context given the strength of influence communities can have on individual decision making.

3.5 Conclusion

The current study found the knowledge and understanding of cancer screening for people with learning disabilities and/or autism to be tentative, with uncertainty around applicability, roles in facilitation and decision-making. Access to screening is hampered by systemic barriers of how standard screening programmes are implemented, and at times, the relationships between individuals, family- and paid-carers and healthcare workers. The quality of relationships, including within the wider community, can be a factor conducive to screening which is under-utilized. Several directions for improving uptake have been identified, including calls to review the standard screening systems for these population groups and proactive support for all stakeholders in supporting decision-making for their dependents.

Chapter 4. Discussion

This thesis sought to understand the current uptake of cancer screening and HPV immunization in the learning disability and autistic populations of Orkney and Shetland, the views and experiences of stakeholders and to establish directions and recommendations for improvements. From the audit described in Chapter 2, it is clear that overall cancer screening and HPV uptake is lower in the learning disability and autism populations than in the general population. Rates are comparable to data on learning disability and autism populations elsewhere in the UK but worse in the case of bowel screening in Orkney and breast screening in Shetland. Chapter 3 offers insight into the views and experiences described by people with learning disabilities and/or autism, family guardians and health and social care workers as to why this might be. High-level structural barriers to screening, including how screening and wider health and social care systems function, uncertainty around roles, responsibilities and decision-making, as well as how cancer, screening and people with learning disabilities and/or autism are viewed in the community. While small communities were seen as a unique strength of remote and rural islands, they were also felt to be a factor in perpetuating unhelpful myths and stereotypes.

This final chapter will provide some direction and recommendations to improve the uptake of screening and HPV programmes in Orkney and Shetland. An initial co-production approach to creating a programme of work will be outlined and overall reflections on the research process will be discussed.

4.1 Recommendations

4.1.1 Targeted Efforts

Given the generally high uptake of cancer screening in Orkney and Shetland the figures outlined in the audit are disappointing and concerning, and suggest that whatever factors are at play with promoting uptake in the general populations of the isles do not extend to the minority populations of those with learning disabilities or autism.

Clearly, targeted efforts are required for all programmes, to raise awareness of the disparity and highlight that screening and HPV immunization are programmes which are inclusive of the learning disability and autism populations. One such approach could include community screening events, such as the ScreenABLE project developed through the University of Illinois, Chicago (Magasi et al., 2019). Through a collaboration with community partners and women with disabilities, they developed a short film to educate and raise awareness of breast cancer disparities and barriers to screening. In addition, they established a wellness fair, which featured accessible mammograms, interactive demonstrations and workshops highlighting the importance of preventative, health-promoting behaviours such as exercise and nutrition. While long-term evaluation of the ScreenABLE project remains to be seen, it showed promise through increasing interest and attendance in its second year.

It has been suggested that participation in one programme could be used to promote participation in others (Kotzur et al., 2020; Labeit & Peinemann, 2015; Scott et al., 2021). Given that the majority of women eligible for all three screening programmes in the current study participated in at least one, there is scope for awareness raising activities or community screening fairs for women to cover all three programmes. This may be particularly helpful to identify and extrapolate supportive factors from one programme to another. In addition, each screening programme could act as an educational platform for ‘teachable moments’ (see Lawson & Flocke, 2009) to aid the promotion of others or cancer screening in general, through simply asking about participation in other programmes, and encouraging individuals to consider engaging.

4.1.2 Consistency in Approach

From a Primary Care perspective, there was considerable Practice variability with regards to the follow up of non-attenders, whether screening programmes are discussed in consultations, as well as the processes around making reasonable adjustments. A consistent approach across Primary Care would improve equity of treatment and help reduce uncertainty and confusion around applicability of screening. GP Practices are in a unique position to identify those individuals who are not participating in cancer screening and their general patterns of screening behaviour. The high esteem in which GP’s seem typically held by people with learning disabilities and their family carers mean they are likely to have the greatest influence on the decisions these individuals make on cancer screening. Individuals who do not have familial support or access support services are of particular concern, and their GP may be the only point of contact.

The descriptions of reasonable adjustments made by Primary Care GP Practices and healthcare staff highlight a lack of consistency and in some cases a lack of confidence or conviction with what adjustments can be made. Efforts should be made to formalise reasonable adjustments as standard throughout Primary Care, incorporating for example, tools and training to help build awareness and confidence around proactively implementing reasonable adjustments, rather expecting patients to ask for adjustments to be made.

Given that a multitude of individuals are often involved in the care and support of individuals with learning disabilities and/or autism, there is scope to improve communication and multi-disciplinary working between those involved, which could improve access and education for all.

4.1.3 Data Collection and Recording

The discrepancy in population data numbers between those gathered from Primary Care and those published by the SCLD were higher than expected, particularly for Orkney. There are significant challenges with data sharing between Health Boards and Local Authorities, however it would be sensible to cross-reference the two datasets to identify the extent of the discrepancy. These figures may represent individuals who could benefit from extra support but are not accessing services.

In addition, the audit showed a significant gender disparity in low numbers of older women with learning disabilities in Shetland, which may be indicative of more severe inequalities. It is recommend that both Shetland and Orkney continue to monitor the uptake of cancer screening in these populations as standard to see if these patterns hold with time. It is worth noting that the present audit represents comprehensive baseline data, which can henceforth be compared further to the implementation of any recommendations or interventions.

For data collection to support improved uptake of cancer screening there needs to be improvements in processes and recording. In particular, ensuring consistency in standardized read codes or templates may be helpful for recording learning disability and/or autism diagnoses and evidencing conversations and reasonable adjustments. An in-depth discussion around the pros and cons of recording diagnoses are beyond the scope of the current study, however, the benefits here are clear for ensuring patients receive the best care possible.

4.1.4 Support for Decision Making

One of the most salient issues identified through the qualitative study was the difficulties all stakeholders had in navigating decision making around cancer screening, whether this was a health or social care professional supporting the decision-making process, a family guardian considering decisions with a dependent, or an individual exploring their options. Shared decision making is a key component of the Realistic Medicine approach, which aims to delivering better value care for patients and health and care systems (Scottish Government, 2023). Support for decision making is recognized as integral to Realistic Medicine, however the interventions promoted through this initiative remain fairly high level and still require patients to be more proactive. Supported decision making would aid the challenging juggle of rights, risks and practicalities for both family guardians and health and social care workers and promote the best outcomes for service users (Bigby et al., 2019).

Motivational Interviewing training could provide health and social care staff with foundational skills to navigate difficult conversations (Chan & So, 2021) and training which highlights the pervasive stigma and myths around people with learning disabilities and autistic people is arguably essential. The La Trobe Decision Making Framework provides an example of a novel, evidence-based resource for engaging in effective support (Bigby et al., 2022; Douglas & Bigby, 2020). The La Trobe resource emphasizes the will and preferences of the individual, as well as highlighting how decision making always takes place within a relational context, which can be shaped and influenced by the beliefs and values of supporters.

4.1.5 Enabling Screening

Finally, there feels to be a gap in education and support for health and social care staff, and possibly family guardians, to enable screening, should an individual decide they would like to go ahead. Tools and training for helping an individual through a screening process, such as relaxation training, could be extremely helpful. In addition, acknowledging the difficult feelings within individuals is an important step, while also then trying to find ways to inject humour and fun into the process. Efforts should be made to make the first screening experience as positive as possible, as this may influence the likelihood of the individual undergoing subsequent screenings.

4.2 A Co-Production Plan

Co-production is a complex concept which has no single definition. The Social Care Institute for Excellence (SCIE, 2023) highlights key features of co-production initiatives, including: those who access care and support are seen as people with skills; barriers are broken down between people who access services and professionals; people’s existing capabilities are built upon; reciprocity and mutuality; peer and personal support networks work alongside professional networks; organisations become agents for change, as opposed to purely service providers. Co-production offers principles for a collaborative approach to screening, which in its essence, could support the consistent, targeted and multi-stakeholder foundations recommended above. Step-by-step guides are not advised for co-production projects, as each project will require different approaches in order to be effective. The SCIE however promotes a jigsaw model (see figure 4) to support considerations for implementing co-production projects. Appendix 9 outlines action points in response to recommendations under the four jigsaw ‘pieces’ of culture, structure, practice and review.

Figure 4. Jigsaw model of co-production considerations

Note: Source https://www.scie.org.uk/co-production/what-how

4.3 Strengths and Weaknesses

One of the main strengths of the current thesis as a whole includes taking an academic research approach in a practice-based setting. Whilst the delivery of a project in academic settings is in many ways incongruous to delivery in an NHS setting, there are some significant advantages to working in this way. Firstly, the process is significantly slowed through ethical processes and appropriate protocols. While this frustrates progress, time is allowed for more careful reflection and deliberation of the minutiae of the project, highlighting factors which may have been otherwise overlooked. For example, it may have been all too easy to deliver a speedy project implementing a new ‘tool’ of sorts, or a generic awareness-raising campaign. However, this would not have even scratched the surface of the underlying systemic barriers described by participants. The majority of quick-turnaround projects would be at best short-lived, and at worst ineffectual and wasteful. In addition, the ethical process in itself ensures that those involved have adequate safeguarding and provides confidence during implementation.

A further strength of taking a research-based approach is drawing from a canon of previous literature and research in a variety of settings. This allows for more lateral and creative thinking, and a depth of argument which could be the marker of success against fierce competition in future funding bids.

Another strength of the current study has been to access the unique knowledge and perspectives of those local to the northern isles. While the information gleaned highlights that many of the issues are the same as described nationally, engaging with stakeholders at a local level allows for a tailor-made approach, reduces wasted resources and allows for a genuinely person- and community-centred approach.

One significant weaknesses of the project has been the combining of the learning disability and autism populations. The reasons behind this are explained in Chapter 1(1.5), however even from the participants of the current study it was clear that these populations are distinct and the snapshot of quotes obtained are not sufficient to describe the diverse views likely to exist. Further, participants in the current study were all broadly open and supportive of cancer screening, therefore the views of those who consistently decide not to participate are missing and should be a focus for future research.

4.4 Research Reflections

The context of Covid-19 pandemic, in which the current study was conducted presented some unique challenges worth noting. Most significant were the restrictions and delays, which made the recruitment of focus groups much more challenging. While the project was initially conceptualized as comprising of mostly focus groups, the majority of participants chose to engage on a one-to-one basis. The dynamic in focus groups is of course different and it is worth considering whether individual’s narratives or responses would be more or less authentic alone, or in the presence of and in conversation with peers.

The partnership working between two health boards was also hindered by the pandemic, such that Public Health resources were often diverted to contact tracing efforts and therefore momentum was hard to maintain. The most challenging aspects of joint-board working include competing agendas, which was compounded by the project staff turnover: those who initially conceptualized the project were no longer involved, leaving others to renegotiate arrangements in a completely alien context. Accessing data from another Board also presented significant difficulties, with different processes making it harder to know where to look and who to ask, and further challenges in delegating responsibility. Despite these issues, the joint project allowed a clear sense of camaraderie between the two Public Health Teams, and a strong satisfaction can be gained from sharing resources, recognising reflections of each island community in the other, and a shared fortitude from seeing through the project whilst enduring the pandemic.

4.5 Conclusion

This research is the first to explore cancer screening, specifically in the learning disability and autism populations, of any of the more remote and rural populations in Scotland. The rates of screening uptake are comparable to available data on the learning disabilities populations elsewhere in Scotland, but serve to highlight a greater disparity in Orkney and Shetland, given the relatively high uptake of screening in their respective general populations. Targeted efforts are essential to redress these inequities, for which we now have evidence-based recommendations and a co-production plan.

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Appendix 1. Ethical Approval Letter

University of Stirling
Cottrell 3B1
Stirling
FK9 4LA

04/10/2021

Dear Astryd

Ethics Application Form : Cancer Screening for People with Learning Disabilities and Autism 1971

Thank you for your submission of the above ethics application.

The ethical approaches of this project have been approved and you can now proceed with your project.

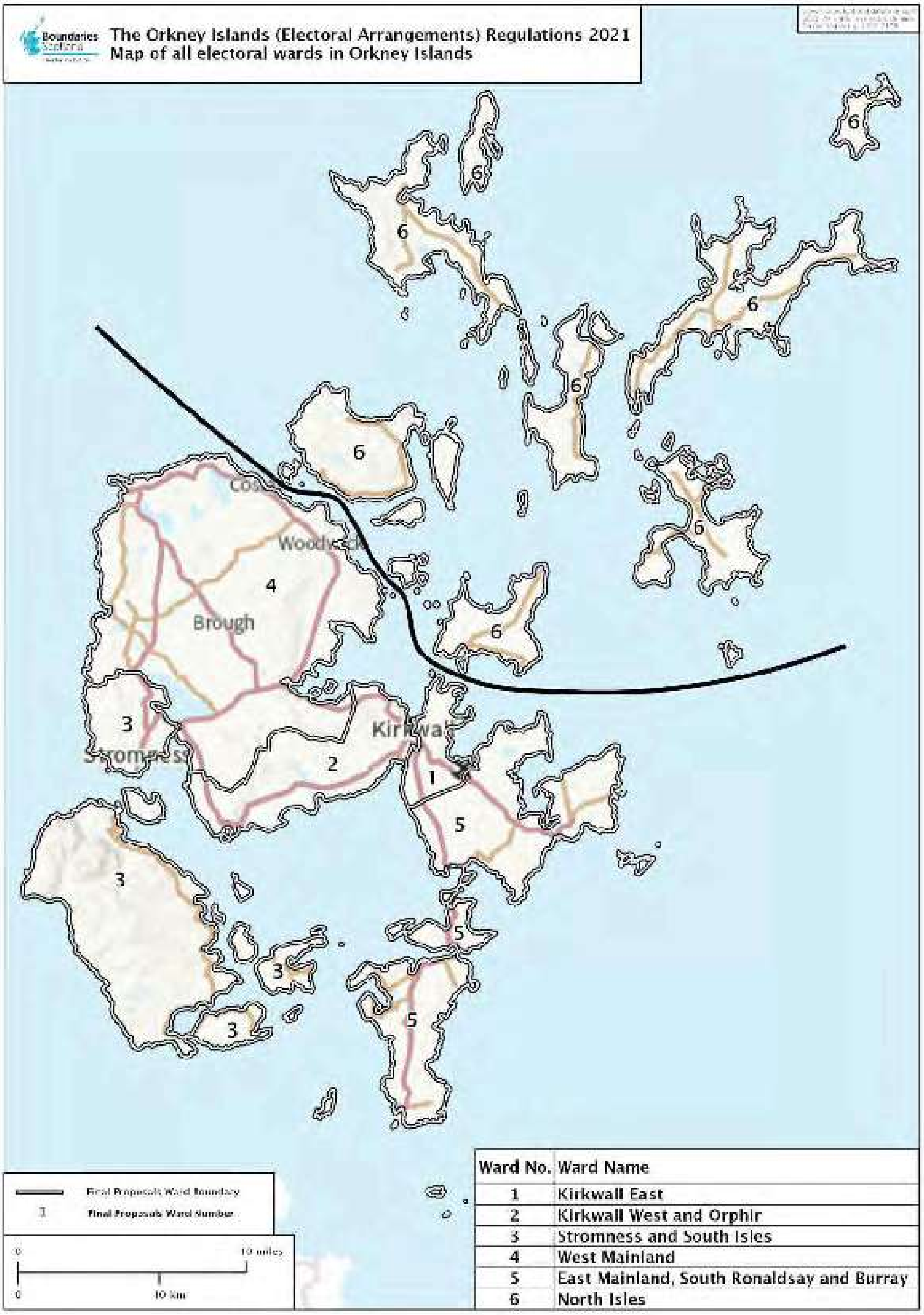
Please note that should any of your proposal change, a further amendment submission will be necessary.

If you have any further queries, please do not hesitate to contact the Panel by email to ethics@stir.ac.uk

Yours sincerely,

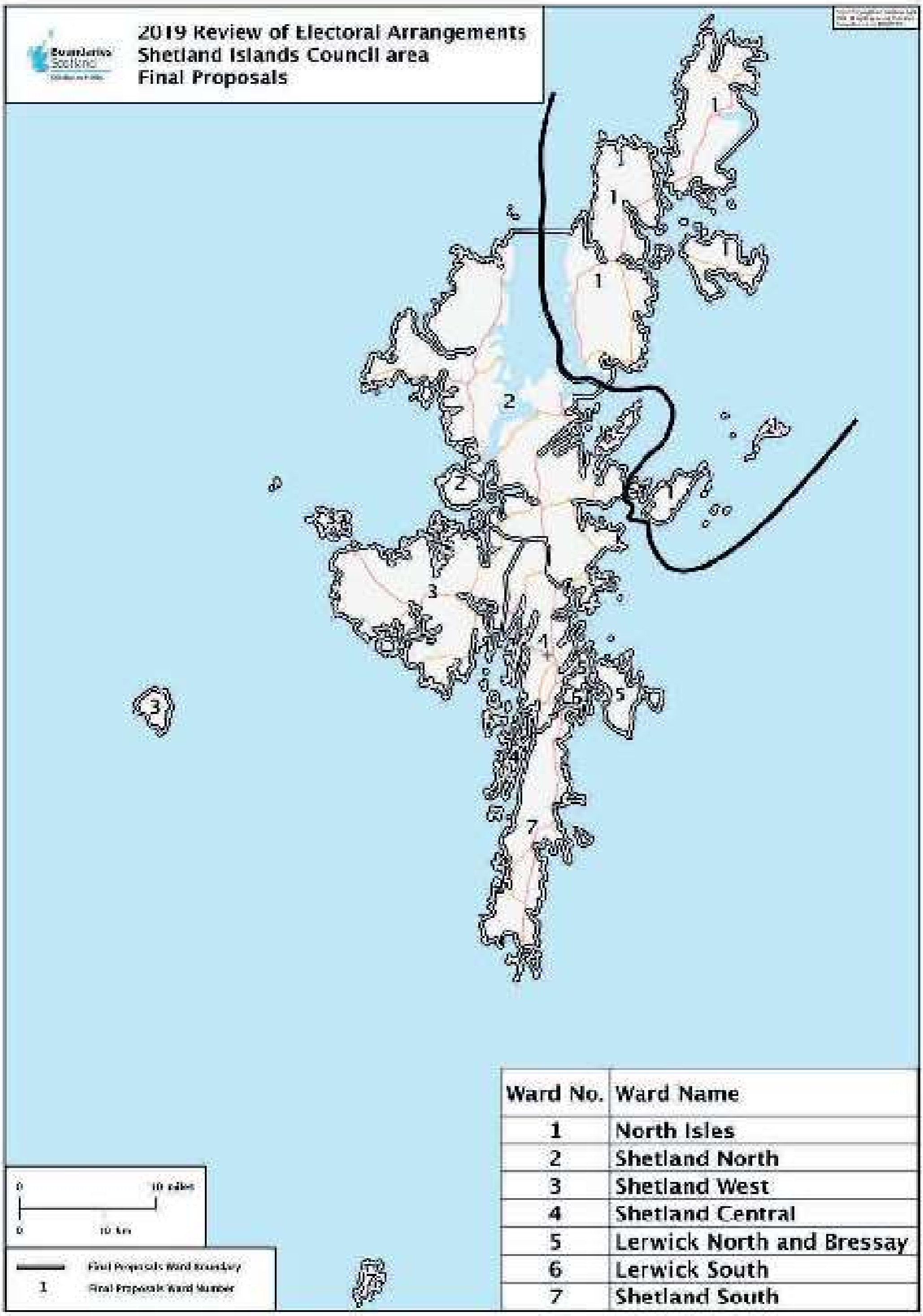
NHS, Invasive or Clinical Research

Appendix 2. Orkney Electoral District Map



Ward No.	Ward Name
1	Kirkwall East
2	Kirkwall West and Orphir
3	Stromness and South Isles
4	West Mainland
5	East Mainland, South Ronaldsay and Burray
6	North Isles

Appendix 3. Shetland Electoral District Map



Ward No.	Ward Name
1	North Isles
2	Shetland North
3	Shetland West
4	Shetland Central
5	Lerwick North and Bressay
6	Lerwick South
7	Shetland South

Appendix 4. Coding frame categories, subcategories and sub-subcategories and respective definitions.

Main Category	***Sub Category***	**Definition (what is meant – what features)**
Recording Information		Any mention of methods, processes or protocols used to formally retain information about the patient or relating to their care. May include diagnoses, refer to certain codes or templates or locations where this information is kept.
	Confirmed Diagnosis	Refers to officially confirmed diagnoses of learning disability or autism from a source of authority.
	EMIS Codes	Refers to the use of "read codes" on the electronic patient record system used in Primary Care (EMIS).
	Local Knowledge	Describes local, community or lay knowledge about individuals.
	Consultation Notes	Describes the use of consultation, or medical notes (on EMIS) as a way to record information.
Reasonable Adjustments		Any mention of reasonable adjustments or adaptations to any aspect of a patient's healthcare experience, and associated processes or protocols, designed to enable patients to access cancer screening or healthcare more generally.
	Uncertainty	Expressions of uncertainty, doubt or lack of knowledge around what reasonable adjustments are, or how they are made within a Practice.
	Alerts	Refers to mentions of using the alert system on EMIS to highlight a support need or a requirement for reasonable adjustments.
	Accommodating	Refers to adapting usual course or process of care to enable an individual to access healthcare.
	Flexibility	Refers to prioritizing the needs of the individual, and making allowances to meet these through practical adjustments.
	Time	Refers specifically to allowing additional time, creating space in diaries or retaining appointments at specific times for individuals.
	Information	Refers to the provision of information or education. May refer to conversations, phone calls, leaflets, written summaries or other to aid knowledge and understanding.
	Guardians	Refers to the including or involving an individual's family carer or guardian in their healthcare.
Follow-up of defaulters		Reference to communication with individuals who have not participated in any of the cancer screening programmes (bowel, breast or cervical) about their lack of participation.
	Letters	Refers to sending letters from the Practice only (NOT letters which are sent from the national screening systems).
	External Service	Reference to the national screening systems as outwith the Primary Care system.
	Encouragement	Refers to communicating with any patients to discuss their not participating in a cancer screening programme and promoting their participation. May be via phone or in person.

Appendix 5. Examples of media advertisements for recruitment

NHS Shetland and NHS Orkney are working together to make **cancer screening** (breast, bowel & cervical) better for people with learning disabilities and autism.

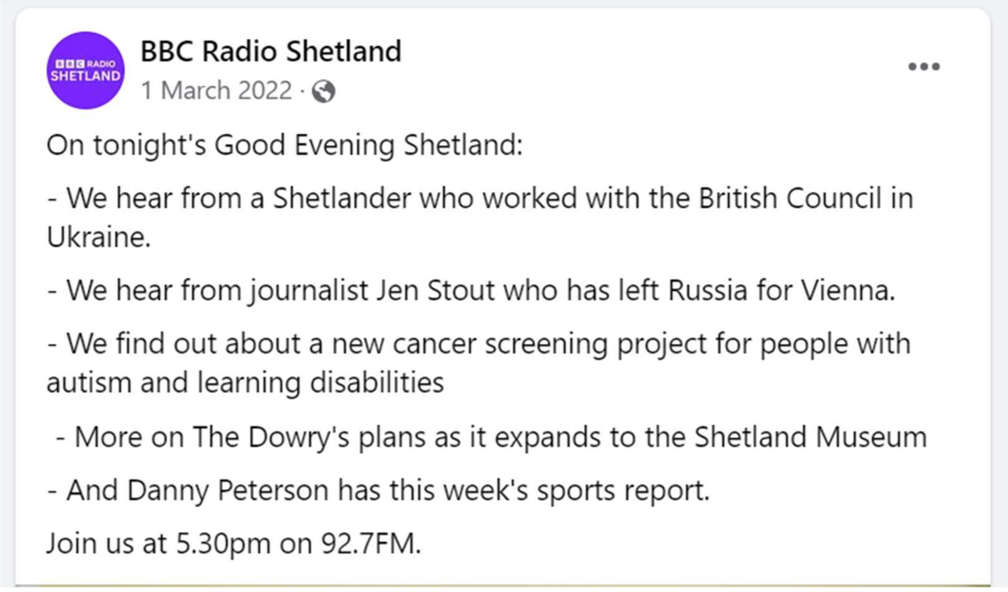
People with learning disabilities or autism
Family members
Carers
Health and Social Care Staff
(in Shetland and Orkney)

We need to hear your views!

If you are interested in helping us or to learn more please contact:

Astryd (Shetland) ✉ astryd.jamieson@nhs.scot ☎ 01595 807494
Jocelyn (Orkney) ✉ jocelyn.strawson@nhs.scot ☎ 01856 888084

You can speak to us face to face, over the phone or online.
All contacts will follow current covid-19 guidance.



BBC Radio Shetland
1 March 2022 ·

On tonight's Good Evening Shetland:

- We hear from a Shetlander who worked with the British Council in Ukraine.
- We hear from journalist Jen Stout who has left Russia for Vienna.
- We find out about a new cancer screening project for people with autism and learning disabilities
- More on The Dowry's plans as it expands to the Shetland Museum
- And Danny Peterson has this week's sports report.

Join us at 5.30pm on 92.7FM.

Appendix 6. British Psychological Society's Capacity Checklist

ASSESSING CAPACITY TO CONSENT TO PARTICIPATION IN RESEARCH - BRITISH PSYCHOLOGICAL SOCIETY

Name of potential participant:

Checklist for researchers to decide whether a prospective participant has the capacity to consent to their participation	
Section A – Enabling capacity:	
Have you made every effort to enable a prospective participant to make the decision themselves to participate or refuse?	☐
Have you used language or methods of communication that the person is most likely to understand?	☐
Have you given sufficient time for the person to think about the project?	☐
Has the person conferred with others who could help explain the project?	☐
If NO to any item in Section A, return to BPS guidance on enabling decision making. **If YES to all items in Section A continue…**	
Section B – Diagnostic assessment	
Is there evidence to demonstrate impairment of mind or brain?	☐
Is there evidence to demonstrate that this is temporary, fluctuating or permanent?	☐
Is there evidence to demonstrate that the impairment affects the person's ability to decide about their participation in research?	☐
If YES to any item in Section B discuss with Principal Researcher. If no to all items in Section B, continue…	
Section C – Functional assessment	
Does the person understand that they can consent or refuse to participate in research?	☐
Does the person understand what the research is about?	☐
	☐

Does the person understand and weigh up the benefits and risks of agreeing or refusing to take part? Has the person communicated their decision to you in any way?	☐
If YES to any item in Section C, return to BPS guidance on 'enabling decision making'. **If NO to the first three items in Section C – the person does not have the capacity to consent to or to refuse to take part in the research project.**	
Checklist completed by: **Date:**	

British Psychological Society (2008) Conducting research with people not have the capacity to consent to their participation. A practical guide for researchers.

Appendix 7. Interview/Focus Group Schedules

Learning Disabled/Autistic People

Bowel cancer screening (Men 50+)

- What do you know about bowel cancer?
    - If no one has heard of it present FIT test to show what it would involve
    - *Has anyone ever spoken to you about bowel cancer?*
    - *Who would you want to talk to you about bowel cancer?*

- What do you know about bowel cancer screening?
    - *How do you find out about bowel cancer screening?*
    - *How do you do a bowel screening?*
    - *What do you think happens after you've done a screening?*

- Has anyone done a screening and would you like to share your experience?

- How do you decide whether or not to do bowel cancer screening?
    - *How do you feel about doing bowel cancer screening?*
    - *What puts you off or stops you from doing a test?*
    - *What would help encourage you to do a test?*

- What would make it a better experience?
    - *RE invitation*
    - *Testing*
    - *Results*
    - *What do other people need to do to help?*

- What questions do you have about bowel cancer screening?

- Is there anything else that comes to mind about cancer screening you'd like to talk about?

Cervical cancer screening/HPV imm (Women 18-26)

- What do you know about cervical cancer?
    - If no one has heard of it can show speculum and swab and discuss what it would involve
    - *Has anyone ever spoken to you about cervical cancer?*
    - *Who would you want to talk to you about cervical cancer?*

- What do you know about cervical cancer screening?

  - *How do you find out about cervical screening?*
  - *What happens when you go for a cervical screening?*
  - *What do you think happens after you've had a cervical screening?*

- Has anyone gone for a cervical screening and would you like to share your experience?

- How do you decide whether or not to go for cervical cancer screening?
  - *How do you feel about going for cervical cancer screening?*
  - *What puts you off or stops you from going?*
  - *What would help encourage you to go?*

- What would make it a better experience?
  - *RE invitation*
  - *Procedure*
  - *Results*
  - *What do other people need to do to help?*

- What questions do you have about cervical cancer screening?

- Is there anything else that comes to mind about cancer screening you'd like to talk about?

Breast, bowel and cervical cancer screening (Women 25+)

- What do you know about breast cancer?
  - If no one has heard of it show images of procedure and discuss
  - *Has anyone ever spoken to you about breast cancer?*
  - *Who would you want to talk to you about breast cancer?*

- What do you know about breast cancer screening?
  - *How do you find out about breast screening?*
  - *What happens when you go for a breast screening?*
  - *What do you think happens after you've had a breast screening?*

- Has anyone gone for a breast screening and would you like to share your experience?

- How do you decide whether or not to go for breast screening?
  - *How do you feel about going for breast screening?*
  - *What puts you off or stops you from going?*
  - *What would help encourage you to go?*

- What would make it a better experience?
  - *RE invitation*

  - *Procedure*
  - *Results*
  - *What do other people need to do to help?*

- What questions do you have about breast screening?

- Is there anything else that comes to mind about cancer screening you'd like to talk about?

Repeat above questions for bowel/cervical - over more than one session if need more time

<u>Family/Unpaid Carer Groups (depending on age of family member with LD/autism include or exclude HPV immunisation)</u>

- What do you know about cancer screening/immunisation programmes?
  - *Have FIT test/swabs/images for prompts if nobody knows about it*
  - *How do you find out about cancer screening/immunisation?*
  - *Who would you want to speak to your family member about cancer and cancer screening/immunisation?*
  - *Have you spoken to your family member about cancer screening/immunisation?*

- How do you feel about your family member attending screening/doing a screening test/receiving HPV immunisation?
  - *Is this different from how you feel about attending screening/doing a screening yourself? Why?*
  - *How have your own experiences with screening shaped how you feel about your family member attending screening/doing a screening test?*

- How do you and your family member make decisions about whether to attend screening or not?

- What needs to be taken into consideration for someone with a learning disability or autism participating in screening programmes/immunisation?

- What do other people need to do to support your family member to participate in screening programmes/immunisation?

- What questions do you have about screening programmes/immunisation?

- Is there anything else that comes to mind about cancer screening/immunisation you'd like to talk about?

Paid Carers

- What do you know about cancer screening programmes?
  - *Have FIT test/swabs/images for prompts if nobody knows about it*
  - *How do you find out about cancer screening?*
  - *Who do you think should speak to the people you support about cancer and cancer screening?*
  - *Have you spoken to the people you support about cancer/screening?*

- How do you feel about the people you support attending screening/doing a screening test?
  - *Have you ever supported anyone to participate?*
  - *What do you think your role is around supporting people with LD/autism to participate in screening?*
  - *How have your own experiences with screening shaped how you feel about this?*

- In your experience, how are decisions made about whether the people you support attend screening/do a screening test?

- What needs to be taken into consideration for someone with a learning disability or autism participating in screening programmes?

- What do other people need to do to support people with learning disabilities/autism to participate in screening programmes?

- Is there anything else that comes to mind about cancer screening you'd like to talk about?

NHS Staff

- How are you involved in cancer screening/immunisation programmes?

- What is your experience of supporting people with learning disabilities/autism in participating in cancer screening/HPV immunisation?
  - *Do you do anything differently for people with LD/autism?*

- What do you feel your role is around supporting people with LD or autism to participate in cancer screening/HPV immunisation?

- What needs to be taken into consideration for someone with a learning disability or autism participating in screening programmes?

- What do other people need to do to support people with learning disabilities/autism to participate in screening programmes?

- What questions do you have around supporting people with learning disabilities/autism with cancer screening?

- Is there anything else that comes to mind about cancer screening you'd like to talk about?

Appendix 8. Theme development from within group analysis

Service users	
Theme	**Description**
Screening or cancer	Cancer screening, for those participants who had been through with it, is seen as a good, or more strongly, an essential thing, but against an almost inevitable alternative of cancer and potential death, described in very black and white terms. Included in this is the societal discourse on cancer and cancer screening, individuals' friends and family experiences of cancer and their deaths, what they hear in the media and their other experiences of healthcare more generally.
Screening is something you have to endure	The entire process of cancer screening, from navigating the system, experiencing the test and waiting for results, requires tolerating a spectrum of distress, ranging from mild nuisance and discomfort through to real fear and significant pain. Generally, screening is tolerated and patients highlight the speed and ease, and resulting relief and reassurance from their negative results. At the upper limit of distress associated with screening our human threat-response comes into play. Some dissonance here: participants can objectively recognize difficult feelings and they maintain perceptions of the importance of screening but protect themselves by closing, avoiding or distracting from discussions. Participants relate screening to their own or others' health problems or healthcare experiences they've had to endure, and include some methods or ideas for coping, some of which are more adaptive (e.g. humour, seeking reassurance from others, relaxation) while others are less so (e.g. avoidance, distraction) Extreme measures such as anaesthetic proposed as a solution. Issues of choice and consent are palpable.
Other people are gatekeepers to screening	People with learning disabilities are dependent on their support network for information, communication and understanding screening. This responsibility falls primarily on carers but also includes health professionals (who are specifically responsible for the systems within which screening operates, as well as the tone and accuracy of communication around dissemination of information around screening), and peers (who can provide emotional support and reassurance, particularly if they have been through screening themselves). Service users' discourse are reflective of their disempowerment. Patients defer to the decisions of healthcare professionals and support professionals in a paternalistic system; going against this is rarely considered, or viewed as making trouble.
Family Carers	
Theme	**Description**
The burden of responsibility	There is a heavy burden of responsibility which falls on the family to act as guardians for their family member with autism/learning disability, to manage and make decisions around their holistic health and wellbeing, which includes cancer screening. This is seen as a continuation but also an extension of a parenting role, which is different from mainstream families due to the nature of their person's disabilities and the involvement of professionals. People with learning disabilities or autism are at risk of being missed or overlooked because of difficulties in communicating need, they lack of knowledge and understanding or they lack opportunities to be heard. These

	difficulties evoke fierceness and protectiveness in family carers who have fought long and hard to be heard and for the rights of their child. This demanding and exhausting position brings with it significant concern about who takes on the responsibility of the person's care if or when parents die. The nature of the role of family carers and the dilemma around complex healthcare decisions such as cancer screening amplifies the tension between parenting, acting as a guardian and allowing adult children their independence and right to choice. This becomes more apparent when comparing siblings of the same family who have different levels of need and is particularly salient with cervical screening/HPV immunisations and discussions around sex and relationships. Family carers often describe an authoritative style around healthcare decision making. Parents express awareness of where their feelings conflict with their desire to protect and do what's best for their dependent, such as wanting to protect from discomfort, and recognise the risk of projecting these feelings onto their dependent.
Dependence on healthcare systems and professionals	Family carers express a dependence on medical professionals (primarily their dependent's GP) to raise awareness, communicate about screening and provide advice and guidance in the first instance. Health professionals and particularly doctors are generally well-respected, with valued relationships built on familiarity, continuity, trust and shared decision-making. However, the quality of relationships vary and are dependent on the health professional's attitude and skill. Participants expressed experiences of stigma, feeling unheard, dismissed and responsibility handed back to them despite their lacking the medical knowledge they felt was necessary.
Living in a system	Families with learning disabled or autistic family members who require support live within a ubiquitous system, and are surrounded and managed by multiple professionals, 'just because'. Services are felt to be generally supportive, however participants highlight the bureaucratic nature of the system and the lack of support for parents, who often feel under close scrutiny and left in the dark, outside the system. This is especially so for family carers of adult children, who reflect on the contrast between services for children and young people and the transition into adult services, where they feel they have much less control, despite the continued burden of responsibility. Family members often describe instances of feeling unheard, or ignored, despite their close understanding of the person.
The protection of community	Trusted, quality relationships and support networks including third sector services, activity clubs and employment act as a protective factor around individuals with learning disabilities or autism, which can relieve the burden on family carers and serve to normalize conversations around screening. These support services act as a community, and are seen as nurturing, inclusive and respectful. Staff and carers are praised as being proactive and strength-based, focusing on building confidence and motivation. There remains uncertainty around who is best-placed to take a 'lead' on cancer screening conversations but ultimately a unified approach is essential alongside the need for people with learning disabilities/autism to have quality relationships with experienced, proactive people who know and understand them and will exercise vigilance.

Cancer visibility	Participants express an awareness of cancer, screening and the impact on everyone’s lives, especially through personal experiences of family or friends’ deaths due to cancer, and for this reason cancer screening is felt to be positive and important. The visibility of cancer and how it is discussed in the community is felt to be an important aspect of promoting cancer screening, with several accounts of the participants’ dependents expressing fear or worry around the merest mention of cancer, with a strong association with death, but also anxiety around medical interventions more broadly.
Screening needs careful preparation	Participants describe in-depth the preparation required for their dependent to be able to engage in cancer screening, often comparing to other healthcare experiences, such as going to the dentist. Time, repetition and quality relationships are essential conditions, with a positive, gentle and reassuring approach tailored to the unique needs of each individual, to build confidence and normalize. Information needs to be simple, relevant and accessible – linking to how cancer and screening are discussed in society. A challenge participants describe is the information relating to (breast and cervical) screening is confusing and seen as contradictory to previous lessons on body privacy and consent.
Paid carers	
Themes	**Description**
Tension between respecting decisions and duty of care	Support workers hold compassion for the families they support, describe guardians opting out of screening on behalf of their dependent; their perception is this is based on fear and a desire to protect their family member. The expectations are that the person won’t cope with the screening test or the potential negative outcomes. These decisions are felt to be hasty, without considering the opportunity or even trying, and are thought to be primarily driven by the guardian’s own projected feelings. Support workers describe a conflict between their duty of care to the people they support and respecting the decisions and privacy of the family and guardians. To support workers, the importance of cancer screening is implicit, and raising it as an issue is important, but there is a hesitancy to know how to start the conversation or whose responsibility it is. Family might speak to carers but will often have already made up their mind and are closed to any new information or ideas. Support workers describe a need to flip the narrative so that engaging with screening, as opposed to avoiding it, feels like taking control and protecting their dependent, and note their role in keeping the door open and revisiting screening, while remaining mindful of the importance of maintaining trust, positive relationships and the time required to create change.
Screening needs mainstreamed	The conflict for support workers would feel less if they knew for certain decisions made by guardians about cancer screening were informed. Information needs to be in the public domain, in multiple formats to raise awareness and normalize screening in order to instigate open conversations across society. Seeing information as mainstream can tackle fear. Support workers express some discomfort about discussing cancer screening without some prior knowledge or awareness on service user’s part, and still feel that

	these conversations may be better placed at home in a relaxed environment, for the most part, which is somewhat contradictory to the theme but is likely reflective of the anxiety around damaging relationships with the families. Support workers feel the need to be educated themselves in the first instance to allow them to have confidence in discussing screening, and feeling more able to translate available information or resources to suit individual need.
Health and social care staff as facilitators and barriers to cancer screening	Support workers take a broadly problem-solving stance towards facilitating screening; asking questions and offering solutions throughout. Solutions included incorporating screening discussions into annual reviews, promoting a multidisciplinary approach and focusing their efforts on supporting the family members and guardians in the first instance. Support workers described other healthcare staff as broadly helpful and supportive, and having important roles in promoting screening and engaging in discussions around it, as well as supporting their staff teams more generally. In particular, familiar GPs are valued as they are trusted and know the individual, their history and quirks. Generic health appointments or check-ups are seen as good opportunities to pick up missed screening appointments. The Learning Disability nurse is also recognized as a valuable resource, although there is an awareness of demand on their service which perhaps creates a reluctance to seek support. The close familiarity of service users with health and social care staff provides guardians with confidence that screening is unnecessary as any changes will surely be noticed. This causes great strain and worry in support workers who feel under pressure to remain vigilant. However, the potential for support workers to negatively influence family members based on their own attitudes or projected feelings is also noted.

Health care staff

Themes	**Description**
Support begins and ends at the consulting room door	Healthcare staff working within the NHS are very clear on their roles, and the role they play with screening, but not always on each other's. Roles primarily revolve around enabling, educating and coordinating care to varying degrees of responsibility depending on level of need and the screening programme in question, with cervical being the only screening delivered within Practices, but their input begins and ends at the consultation room door. They operate separately to the National screening system and acknowledge other systems in play beyond this but are also uncertain about how these systems work. Staff are conscious of the need to work together with each other, and other people involved in the person's life, such as guardians and support staff. Time and space were often cited as critical but limited factors required for successful care. This is highly dependent on effective information sharing and the importance of early conversations before coming to the GP Practice. These conversations could even begin in schools, particularly around the need for HPV vaccinations and following cervical screening. Staff are aware of gaps in Primary Care where they lack knowledge around the population and where they could be doing more, as well as Practice variability, and often take a solution-focused approach pondering questions and providing solutions. The newly introduced annual health reviews being one such example. Meeting other gaps in care is otherwise seen as an additional burden on administrative staff.

Informed choice in balance with risk and responsibility	Informed choice and shared decision making are at the forefront of healthcare staff's thinking around screening. There are clear challenges in navigating consent, understanding and distress, particularly for Practice nurses carrying out cervical screening, but the broader discussion explores tensions around the desire to defer or depend on the GP for ultimate decision-making, contrasting with the idea of everyone supporting people with learning disabilities and autism taking responsibility for their wider health and wellbeing, balancing the burden of choice and risk, empowerment and duty of care, together. These ideas feed into the structure of systems, strategies and policies, as well as wider issues around inequality.
Systems and stigma perpetuate inequalities	Healthcare staff are dependent on the screening systems working in the background. The screening systems and procedures as well as wider healthcare systems perpetuate inequalities because people with learning disabilities are expected to follow the same procedures as the general population. While procedures are in place to offer reasonable adjustments, and registers of people who have learning disabilities or autism, there exists considerable Practice variability and information gaps, and individuals who struggle to access mainstream services such as screening may not be known to staff. Particularly vulnerable are those who may be borderline, or who are not in support services and may go unseen. Small communities and local knowledge compensate for these gaps in helping identify those who might otherwise be missed, however the local cultures sometimes feed into other issues, such as perception of doctors, not wanting to be troublesome. Inequality is also fueled by pervasive myths, stigma and culture, compounded by a lack of education and engagement. There is a sense of fear, primarily for getting things wrong. This theme is particularly salient for LD nurses, who feel included in the stigma surrounding people with learning disabilities and experience exclusion and ignorance from other nursing colleagues. This theme is best illustrated through the controversy surrounding cervical screening, where assumptions around sexual activity are often made and acknowledged, and while efforts are made to try to enable screening, these can hit barriers of time and confidence, and ultimately decisions still come down to assumptions of sexual activity, which wouldn't be in question for other members of the general population.
How do we sell screening?	There is a strong awareness across healthcare staff for the need to impart knowledge and education, engage in conversations around screening and multi-disciplinary working to promote the benefits of screening. However, this requires buy-in from those involved and consideration must be given to marketing, or how we communicate with others. For example, professionals need to see the economic benefits and not just the extra time involved; people with learning disabilities and their supporters might engage more fully were enjoyable ways of engaging with screening found.

Appendix 9. SCIE 'jigsaw' model of co-production, recommendations and action plan

Recommendations	Action Plan
Culture • Ensure that co-production runs through the culture of an organisation. • Ensure that this culture is built on a shared understanding of what co-production is, a set of principles for putting the approach into action and the benefits and outcomes that will be achieved with the approach. • Ensure that organisations develop a culture of being risk-aware rather than risk-averse.	As the current project sits within the NHS and includes stakeholders from other organisations, it is recognized that to influence the broader culture throughout these organisations will take time and is beyond present scope. In order to ensure a culture of co-production within the co-production team, we will: • Ensure stakeholders are adequately represented in the co-production team, including those identified as missing in the current study (e.g. men with learning disabilities; social workers) • Provide the team with accessible training on co-production • Agree as a team, on a 'branding' to support a sense of ownership (Leask et al., 2019) • Agree as a team the group rights and responsibilities (Leask et al., 2019) • Ensure the team are aware of and consider potential risks and challenges involved (e.g. long-term nature of change; time investments)
Structure • Involve everyone who will be taking part in co-production from the start. • Value and reward people who take part in the co-production process. • Ensure that there are resources to cover the cost of co-production activities. • Ensure that co-production is supported by a strategy that describes how things are going to be communicated. • Build on existing structures and resources.	• Ensure stakeholders are adequately represented in the co-production team, including those identified as missing in the current study (e.g. men with learning disabilities; social workers) • Consider whether the remaining project budget is sufficient to pay the co-production team for their time, whether further funding is available, or the team can agree alternative rewards. • Agree as a team the group rights and responsibilities (Leask et al., 2019)

	• Agree as a team on the overall strategy using the PRODUCES framework (Leask et al., 2019) • Identify existing strengths, resources and skillsets, and agree how these might best be used as part of the co-production teams aims.
Practice • Ensure that everything in the co-production process is accessible to everyone taking part and nobody is excluded. • Ensure that everyone involved has enough information to take part in co-production and decision making. • Ensure that everyone involved is trained in the principles and values of co- production and is supported in developing any new skills they will need for the work they do. • Ensure that frontline staff are given the opportunity to work using co-production approaches, with time, resources and flexibility. • Provide any support that is necessary to make sure that the community involved has the capacity to be part of the co-production process. • Ensure that policies and procedures promote the commissioning of services that use co-production approaches. • Ensure that there are policies for co-production in the actual process of commissioning.	• Agree on the most appropriate, accessible methods to implement the strategy, e.g. Talking Mats, Concept Mapping, Storytelling (Agnello & Longworth, 2022) • Create accessible participant information guides for taking part • Allow plenty of time and opportunities to ask questions prior to participants agreeing to join the co-production team. • Provide the team with accessible training on co-production • Retain budget to allow for further participant training and/or development • Ensure that all participants are supported by their employers or other relevant supporters by being given the time to and any resources required to take part • Ensure the agreed times and venues for the co-production team meetings are suitable for all, and can be flexible to meet the needs of participants • Offer any support required to facilitate participation (e.g. travel expenses, support workers, adequate breaks) • Consider developing a co-production policy/protocol for within the NHS Board
Review • Conduct regular reviews to ensure that co-production is making a real	• Agree time points for review meetings, where progress is considered against the agreed

difference and that the process is following the agreed principles. • Co-produce reviews and evaluations. • Use the review findings to improve ways of applying the principles of co-production, so that continuous learning is taking place. • During reviews and evaluations, work with people who draw on care and support and carers, to think about ways of showing the impact that co-production has, as well as the processes that are involved.	strategy and the team rights and responsibilities are reviewed • Agree shared authorship of any recordings of meetings, reports and resources • Share the responsibility for recording meetings and delivering evaluations • Agree to create a reflective review of the co-production process to share widely with others • Agree a process of evaluation for the project.