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Thesis submitted in partial fulfilment of the degree of Doctor of Clinical Psychology (DClinPsych)

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Theory Driven Research Project:

How do autistic traits influence affirmative experiences of gender identity?

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Executive summary of Theory Driven Research Project:

658 words

Connecting Narrative:

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Abstracts

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Scoping review: Does Psychoeducation for Non-Epileptic Seizures increase quality of life and/or decrease seizure occurrence?

This research examined the content and efficacy of psychoeducational interventions for non-epileptic seizures (NES) in terms of seizure frequency and quality of life. Databases were searched and seventeen eligible studies identified. Findings revealed that components and modalities of interventions varied greatly, though all included information about NES, its symptomology and the mind-body link. Methods of evaluating the interventions also varied and though reporting on significance of findings was inconsistent, there was a general trend in symptom reduction and improvement in general wellbeing. Findings illustrate that psychoeducation has potential benefit as an intervention in its own right, however inconsistency in the findings suggests that psychoeducation is unlikely to be a sufficient intervention as a standalone treatment for most people and may be best placed as a second step in a stepped care model, between initial diagnosis and further psychological treatment. Furthermore, the present literature needs replication and greater sample sizes for more certain conclusions to be drawn.

Keywords: Epilepsy, Non-epileptic seizures, psychoeducation, psychology

Service Improvement Project: Decision making in NF2 Hearing Interventions

The project aimed to understand experiences of people with Neurofibromatosis type 2 (NF2) under the Oxford and South West service, in terms of psychological impact of NF2 and support required for decisions to take up hearing interventions (hearing aids, auditory brain stem implants (ABI) and cochlear implants (CI)). Fifty surveys were completed and analysed. Qualitative findings demonstrated physical symptoms contributed to emotional

distress, social isolation and challenged sense of identity. Quantitative findings showed people generally felt informed and ready to take up hearing interventions, with many feeling satisfied with the outcome. People without CI/ABI expressed that surgery risk and visibility of devices were barriers. Service recommendations include increasing accessibility of psychological support before interventions, greater provision of information, and consideration of communication and emotional needs of patients.

Key words: Neurofibromatosis, NF2, Hearing, Audiology, Cochlear implant, Hearing aid, Auditory brain stem implant, Neurology, Oncology, Psychology, Adjustment.

Theory Driven Research Project: How do autistic traits influence affirmative experiences of gender identity?

This research examined the experiences of gender diverse people with differing levels of autism spectrum disorder (ASD) traits in their physical and social affirmations of gender. Responses to an online survey were split into two groups and analysed according to high and low ASD trait levels. Findings revealed that no differences existed between the groups in importance and sense of passing as their identified gender. There were also no differences between groups in social belonging to gender diverse groups and gender congruence. The only significant difference found was that people with higher levels of ASD traits reported lower psychological functioning. Thus, services need to be mindful of ensuring they are autism-informed in their standard approach and recognise the additional stress that ASD can place on wellbeing. This is particularly salient when people belong to multiple minority groups where impression management occurs across an intersection of stigmatised identities.

Key words: Transgender, Autism, Affirmation, Social belonging

Scoping review: Does Psychoeducation for Non-Epileptic Seizures increase quality of life and/or decrease seizure occurrence?

Word count: 4694

With thanks to Dr Fiona McFarlane and Lawson Falshaw

Target journal: Seizure- European Journal of Epilepsy

This journal has been chosen due to the topic the review being relevant to the journal as well as the journal welcoming reviews on subjects related to epilepsy and other seizure related disorders. This journal also published literature cited in this review so it is likely that this would fit the scope of papers the journal is interested in.

Abstract

This research examined the content and efficacy of psychoeducational interventions for non-epileptic seizures (NES) in terms of seizure frequency and quality of life. Databases were searched and seventeen eligible studies identified. Findings revealed that components and modalities of interventions varied greatly, though all included information about NES, its symptomology and the mind-body link. Methods of evaluating the interventions also varied and though reporting on significance of findings was inconsistent, there was a general trend in symptom reduction and improvement in general wellbeing. Findings illustrate that psychoeducation has potential benefit as an intervention in its own right, however inconsistency in the findings suggests that psychoeducation is unlikely to be a sufficient intervention as a standalone treatment for most people and may be best placed as a second step in a stepped care model, between initial diagnosis and further psychological treatment. Furthermore, the present literature needs replication and greater sample sizes for more certain conclusions to be drawn.

Keywords: Epilepsy, Non-epileptic seizures, psychoeducation, psychology

Introduction

Non-epileptic seizures (NES) are seizures that are not caused by an electrical disturbance in the brain and instead are understood as a physical reaction to emotional stress (Operto et al., 2019). NES are one of many medically unexplained symptoms and can be referred to by multiple names (Appendix B). It is estimated between 2 and 33 people out of 100,000 experience NES, and 20% of people referred to specialist services with epilepsy have NES (Asadi-Pooya & Sperling, 2015; Mellers, 2005).

Quality of life (QOL) refers to the mental, physical and social impacts on a patient, caused by illness and its associated treatments (Su et al., 2017). Individuals with NES report lower

QOL and higher anxiety and depression scores than those with epilepsy (Flewelling et al., 2020b; Karakis et al., 2014; Robson et al., 2018) as well as a reduction in employment (Reuber, 2008). Other studies also report high levels of psychiatric comorbidities and other medically unexplained symptoms in people with NES relative to the general population (Bowman, 1993; Reuber, 2008). Gagny et al (2021) found co-morbid mental health difficulties, but not frequency of seizures, were associated with poorer QOL in adults with NES, a finding also replicated with adolescents (Flewelling et al., 2020b). Increased QOL scores were associated with greater attendance of mental health consultations (Gagny et al., 2021), suggesting that engagement with mental health services improves QOL in NES populations.

CBT and other psychological approaches have a growing evidence base for treating NES. (e.g., Goldstein 2020). However, communication of NES diagnosis has been reported to be difficult for clinicians (Mayor et al., 2013; Whitehead & Reuber, 2012) and if poorly received, patients can mistrust the diagnosis and disengage from psychological treatments (Howlett et al., 2007; R. Thompson et al., 2009).

There is some evidence suggesting psychoeducational approaches for treatment and communication about NES diagnosis are important in reducing seizures, increasing QOL and increasing engagement in other psychological approaches (Mayor et al., 2012; Wiseman et al., 2016). However, the extent to which they can be seen as a treatment in their own right remains a key question, as does the optimal format and content of such psychoeducation.

This review aims to provide a comprehensive overview of what is known about the impact of psychoeducation on QOL, and seizure outcomes in NES as well as summarising the content of such approaches. Previous systematic reviews have looked at mutliple

treatments for NES including psychoeducation but none have examined the impact across the agespan, on QOL specifically or summarised content of the interventions (Appendix C). Therefore, this review seeks to capture the breadth of approaches to psychoeducation as well as the impact on QOL across populations.

Research Questions:

1. Does psychoeducation improve QOL in people with NES?
2. Is psychoeducation effective in reducing seizure frequency in people with NES?
3. How is psychoeducation for NES delivered and what does it include?

Methods

The current review systematically searched four databases as well as grey literature, aiming to map the current research about psychoeducation as a treatment for NES as well as the content of psychoeducation. Any report of psychoeducation was treated as potentially eligible, regardless of level of evaluation. To ensure that the review captured what is most likely being currently offered, papers published prior to 2002 were excluded.

Protocol

The protocol was uploaded to the Open science framework (https://osf.io/8xj3v/) in February 2022.

Eligibility Criteria

The review followed the scoping review framework (Arksey & O'Malley, 2005). It included peer-reviewed papers and grey literature such as poster presentations and Ph.D. theses. For inclusion, reports had to have been written since 2002, clearly use some form of psychoeducation for NES and include an evaluation or description of the intervention. The intervention could be individual, group, aimed at adults, adolescents or children, be supplementary to diagnosis or as part of the diagnostic procedure and could include the

presence of family, friends or schools in the intervention providing there was data specific to individuals with NES from the intervention. The following were excluded:

- Reviews
- Papers published prior to 2002
- Papers concerning other psychosomatic conditions, irritable bowel syndrome or chronic pain conditions (e.g. fibromyalgia) and not NES.
- Papers not available in English

Information Sources

Four databases (MEDLINE, PsycINFO, Cinahl and Embase) were searched on March 12th 2022 and updated on March 3rd 2023, (when one additional paper met criteria for inclusion). The search strategy was altered to meet the requirements of individual databases and was developed with the support of a librarian and included the PICO terms seen in Table 1. Further details on the search strategy are available in Appendix D. Duplicate records were identified and removed upon importation to Rayyan (Ouzzani et al., 2016) from Endnote. The Rayyan software used was not classed as artificial intelligence at the time of using. Once included reports were identified, their reference lists were searched for further eligible studies, as were the reference lists of other relevant review studies.

Table 1: PICO Terms

PICO Component	Search Terms
Population	("functional neurological disorder*" OR "FND" OR “functional neurological symptom disorder*” OR “functional movement disorder*” OR "functional disorder*" OR "non-epileptic seizure*" OR "non epileptic seizure*" OR "non-epileptic attack disorder*" OR "non epileptic attack disorder*" OR "non epileptic paroxysmal event*” OR "non-epileptic paroxysmal event*" OR "non epileptic event*" OR "pseudoseizure*" OR "pseudo-seizure*" OR "psychogenic seizure*" OR "psychogenic non-epileptic seizure*" OR "psychogenic non epileptic seizure*" OR "functional seizure*" OR "PNES" OR "dissociative seizure*" OR “conversion disorder*” OR “motor disorder*” OR "NEAD”)
Intervention	(psychoed* OR psycho-ed* OR education* OR education* program*)

Selection of Sources

Following the search of the databases, results were downloaded to Endnote and subsequently uploaded to Rayyan, an online platform for multiple researchers to work on reviews (Ouzzani et al., 2016). Duplicate papers were removed and the remaining ones underwent title and abstract screening by reviewers blinded to each other’s decision to determine which papers would be included for full text review. All records were reviewed by RU, and LF screened 20% of papers, with any uncertainties discussed and any unresolved disagreement on papers (n=2) brought forward to full text review (k= 0.99). Full text review was conducted using Excel. LF screened a random sample of 25% of the papers with a Cohen’s kappa of 1.0 being achieved (Cohen, 1960). Studies included did

not undergo quality assessment, as this is not something that is a requirement in scoping reviews due to the nature of needing to capture as many papers as possible (Arksey & O'Malley, 2005).

Data Extraction

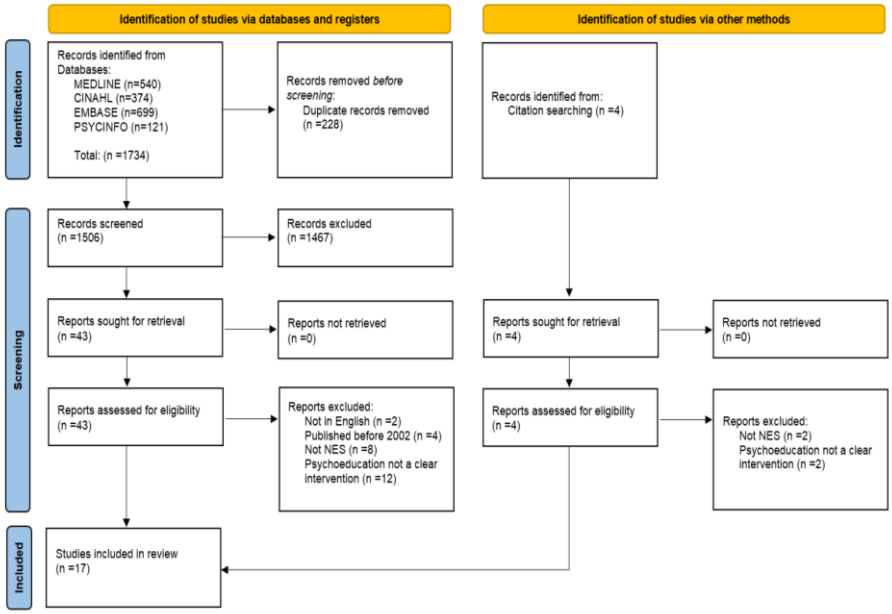
Any outcome data related to the wellbeing, seizure frequency or experience of interventions of participants with NES was extracted from papers and charted using an Excel form. Data was extracted regarding number of participants, location, client group (e.g. adult, adolescent or child), age, ethnicity and sex. The setting, facilitators, length, modality and manualised nature of the intervention was also extracted. Data regarding the content of the psychoeducational interventions was extracted and categorised according to components mentioned in the papers. Where data was not available, or components of psychoeducation not explicitly mentioned, the corresponding field in the data extraction form was left blank.

Results

Selection of Records

Following removal of duplicates, 1612 papers were screened by RU and 323 by LF at the title and abstract stage, with a further 41 papers screened by RU at search update. All texts could be retrieved and were brought forward for full text review (Figure 1).

Figure 1: Record selection in accordance with PRISMA protocols (Page et al., 2021)



Two studies appeared to meet inclusion criteria but were excluded as follows:

1. Novakova et al (2019) did not separate NES from other groups of participants with epilepsy and without NES, therefore the impact of the intervention on NES could not be ascertained.
2. Kozlowska et al (2018) supplemented the intervention with biofeedback training and other inpatient activities, leading to too many confounding factors in the data.

Intervention Characteristics

Interventions had been delivered across five different countries, with the USA (n=8) delivering the majority, followed by the UK (n=6) and Canada, Argentina and Serbia each delivering one intervention. Table 2 presents the characteristics of the included studies.

Format

Most interventions were aimed at adults (n=12), were manualised (n=12) and took a group format (n=10). Three of the manualised groups (adult: n=2, children: n=1)also invited family members or supporters to attend. Three interventions (Flewelling et al., 2020a;

Gurcan et al., 2022; Sawchuk & Buchhalter, 2015) took a family approach whereby the family and patient were seen together in an individual session, all of which were targeted at children and adolescents. Sawchuk & Buchhalter, (2015) also invited the school of the patient to attend this session.

Duration

The timings and durations of interventions varied greatly, with some being single session (n=8), with time allocated ranging from 40-180 minutes, though four of the single session interventions did not specify how long these took. Interventions with multiple sessions ranged from four to ten hours in total, and took place between three and ten weeks, with interventions that had longer sessions tending to be over a smaller time period.

Facilitation and Location

Interventions were led by a variety of health professionals ranging from a consultant neuropsychiatrist (Cope et al., 2021) to an occupational therapist (Wiseman et al., 2016). Five studies had nursing staff delivering the intervention, three were run solely by psychologists and one included a former patient, though five did not report this data. Interventions took place in a range of locations including epilepsy centres (n=3), outpatient clinics (n=4) and lecture theatres (n=2).

Participant Demographics

Participants were mostly female (52%-90% female), in all studies except one which was 73.5% male (Chen et al., 2014). Participants were predominantly white (63.2%-100%) in studies that reported race. Demographic data across studies was limited as some studies did not provide data on sex, race, ethnicity or age of participants, or reported this data before participants had been excluded or dropped out. Participants also had a range of comorbid conditions, with some studies (n=2) excluding people with comorbid epilepsy, and one

excluding any comorbid conditions. However, six studies included people with comorbid epilepsy, and four included people with comorbid PTSD and anxiety.

Table 2: Characteristics of included studies

References	Number of Participants with data at end of study	Location	Client group	Age of participants	Ethnicity	Comorbidity	Sex	Setting of intervention	Who delivers the intervention	Length of intervention	Modality of intervention	Manualised
Zaroff, C. M., Myers, L., Barr, W. B., Luciano, D., & Devinsky, O. (2004).	7	USA	Adult	21 -57*	Not recorded	PTSD, Bipolar II, Substance abuse, Anxiety disorder	6 female, 4 male*	Specialist Epilepsy Centre Outpatient Clinic	Unknown	1 hour weekly sessions for 10 weeks	Group	Yes
Wiseman, H., Mousa, S., Howlett, S., & Reuber, M. (2016).	25	UK	Adult	Mean = 41.8	Not recorded	Epilepsy (comorbid in 2 patients)	52% female, 48% male	Psychotherapy Outpatient Clinic	Epilepsy Nurses, Occupational Therapist, Assistant Psychologists	1 hour weekly sessions for 4 weeks	Group	Yes
Thompson, N. C., Osorio, I., & Hunter, E. E. (2005).	48	USA	Not specified	Not recorded	Not recorded	Not recorded	Not recorded	Epilepsy Centre	Epileptologist	Single session explaining diagnosis with reinforcement of information in follow up by Psychiatric Nurse	Individual	Yes
Thompson, N., Connelly, L., Peltzer, J., Nowack, W. J., Hamera, E., & Hunter, E. E. (2013).	19	USA	Adult	18-66 (mean = 33)	Caucasian 85.7% African American 9.5% Hispanic 4.8%	Exclusion for study	60% female, 40% male	Medical Centre Outpatient Clinic	Clinical Nurse Specialist	40 -90 minute single session	Individual	No

Sawchuk, T., & Buchhalter, J. (2015).	29	Canada	Children and adolescents	90% "adolescent"	Caucasian 79%. Other ethnicities: Middle Eastern, Indian, or Aboriginal descent, no figures given	Epilepsy (comorbid in 24%), other physical health condition (69%). 52% depression, 21% current or historical anxiety disorder. 38% attention, speech, or learning disorder. One patient had been diagnosed with borderline personality disorder prior to the onset of PNES events.	76% female, 24% male	Hospital Outpatient Clinic	Psychologist	Up to three sessions following initial consultation session	Individual with Family and school	No
Sarudiansky, M., Korman, G. P., Lanzillotti, A. I., Pico, M. M. A., Tenreyro, C., Paolasini, G. V., ... & Myers, L. (2020).	12	Argentina	Adult	18-57 (mean= 30.75)	Not recorded	Epilepsy (2 patients), PTSD (6 patients)	83.3% female, 16.7% male	Hospital Outpatient Clinic	Psychologist	Three, bi-monthly two hour sessions	Group with supporters present	Yes
Mayor, R., Brown, R. J., Cock, H., House, A., Howlett, S., Smith, P., & Reuber, M. (2013).	13	UK	Adult	23-38 (mean= 37)*	Not recorded	Not recorded	76 % female, 24% male*	Psychotherapy Outpatient Clinic	Epilepsy Nurses and Assistant Psychologist	Four weekly one-hour sessions	Group	Yes
Libbon, R., Gadbaw, J., Watson, M., Rothberg, B., Sillau, S., Heru, A., & Strom, L. (2019).	97	USA	Adult	Unclear due to reporting of demographics for multiple groups in one	Not recorded	Unclear due to reporting of demographics for multiple groups in one	Unclear due to reporting of demographics for multiple groups in one	Psychotherapy Outpatient Clinic	Unknown	Five weekly 2 hour sessions	Group	Yes

Flewelling, K. D., Koehler, A., Shaffer, J., & Dill, E. J. (2020a).	19	USA	Children and adolescents	9 to 17 years (m = 13.95)	63.2% White, 32% Hispanic or Latino, 26.3% "other," 5.3% Alaskan Native or American Indian, and 5.3% "more than one race"	47% had comorbid epilepsy	90% female, 10% male	Hospital Outpatient Clinc	Multi-disciplinary Team	Single session explaining diagnosis	Family	No
Cope, S. R., Smith, J. G., King, T., & Agrawal, N. (2017).	19	UK	Adult	18-25 (20%), 26-35 (32%), 36-45 (32%), 46+ (16%)	Not recorded	36% had comorbid epilepsy	84% female, 16% male	Neuropsychiatry Clinic	Neuropsychiatrist and Clinical Psychologist	Three 90-minute sessions	Group	Yes
Cope, S. R., Smith, J. G., Edwards, M. J., Holt, K., & Agrawal, N. (2021).	193	UK	Adult	18-25 (16.7%), 26-35 (20.8%), 36-45 (21.9%), 46-55 (24.0%), 56-65 (11.5%). 66+(5.2%)	White (77.9%), Black (7.4%), Asian (7.4%), Mixed/ other (7.4%)	Not recorded	78.8% female, 21.2% male	Lecture Theatre, context of location unknown	Consultant Neuropsychiatrist, Clinical Psychological, Neurophysiotherapist, former patient	One, 1 hour 45 minute session	Group with relatives present (relatives not included in this summary)	Yes

Chen, D. K., Maheshwari, A., Franks, R., Trolley, G. C., Robinson, J. S., & Hrachovy, R. A. (2014).	43	USA	Adult	Mean age of intervention group: 50.76, control group: 50.70*	Not recorded	PTSD, concurrent epilepsy was an exclusion criteria	Intervention group: 26.5% female, 73.5% male. Control group: 23.3% female, 76.7% male (includes later excluded participants)	Lecture Theatre in Medical Centre	Neurologist or Neurology Nurse Practitioner with experience of working with PNES	Three, monthly 1 hour 30 minute sessions	Group	Yes
Baxter, S., Mayor, R., Baird, W., Brown, R., Cock, H., Howlett, S., ... & Reuber, M. (2012).	12	UK	Adult	19- 54	White European (100%)	Concurrent epilepsy was an exclusion criteria. No other data	66.6% female, 33.3% male	Face to face, location unknown	Epilepsy Nurses and Clinical Research Assistant	Four weekly one-hour sessions	Group	Yes
Arain, A. M., Hamadani, A. M., Islam, S., & Abou-Khalil, B. W. (2007).	48	USA	Adult	20-81 (mean= 39)	Not recorded	Not recorded	62.5% female, 37.5% male	Epilepsy Centre	Unknown	Single session explaining diagnosis	Individual	No

Farias, S. T., Thieman, C., & Alsaadi, T. M. (2003).	32	USA	Adult	Mean age of intervention group: 40.36, control group (with confirmed epilepsy): 37.10	Not recorded	Intervention group: depression, anxiety, personality disorder, substance abuse. Control group: depression and substance abuse	Intervention group: 63% female, 37% male. Control group: 40% female, 60% male	Epilepsy Centre	Unknown	Single session explaining diagnosis	Individual	Yes
Vojvodic, N., Ristic, A., Sokic, D., Jankovic, S., & Sindjelic, A. (2006, January).	73	Serbia	Adolescents and Adult	16-57 years	Not recorded	Not recorded	Not recorded	Institute of Neurology	Unknown	Single session explaining diagnosis	Individual	No

Gurcan, L., McAllister, E., Gilmour, J., Green, D. J., McFarlane, F. A., Hadji-Michael, M., ... & Stark, D. (2022).	15	UK	Children and adolescents	11-19 years	Not recorded	40% comorbid epilepsy, 66.7% neurodevelopmental disorders, 33.3% ASD/ASD traits, 13.3% ADHD, 6.7% Tourette's, 7% dyslexia, 7% dyscalculia, 85.7% mental health comorbidities, 33.3% depression, 85.7% anxiety disorders, 53.3% generalized anxiety, 20% social anxiety, 6.7% panic, 6.7% specific phobias, 6.7% OCD, 6.7% PTSD, 40% Physical health comorbidities, 26.7% previous self-harm, 6.7% recurrent headache, 6.7% Recurrent vomiting	66.7% female, 33.3% male	Paediatric hospital	Clinical Psychologists, Assistant Psychologist and Trainee Clinical Psychologist.	Single session, 3 hours	Group with relatives present (relatives not included in this summary)	Yes

**Data not reflective of final sample (e.g. taken before exclusion or drop out)*

Intervention Components

Interventions all covered a general discussion of NES, its symptoms and the mind-body connection (n=17), with one also including a discussion on the different names for NES (Cope et al., 2021) and another discussing names as an ice-breaker (Gurcan et al., 2022), though interventions varied in what other components were included (Table 3). Seven interventions explicitly emphasised the realness of symptoms and that the patient is not "crazy", four conveyed the diagnosis as being good news, and four also conveyed the hope that seizures would resolve over time. One study also had a former patient discussing how they had overcome NES (Cope et al., 2021). Trigger discrimination (n=8), problem solving (n=3), coping with stress (n=7) and relaxation skills (N=10) were taught in interventions. Addressing avoidance behaviours was covered in four interventions, though thought challenge was only covered in one. Interventions also covered QOL and values (n=5) and the need to take responsibility over seizures (n=5). Predisposing factors were discussed in four interventions; some also covered depression and anxiety (n=2), stigma and shame (n=2) and the influence of trauma and abuse (n=2). One intervention discussed the influence of anger (Zaroff et al., 2004) and two discussed understanding emotions generally (Gurcan et al., 2022; Sarudiansky et al., 2020). Two interventions included safety information, though in one this seemed to be aimed more at the individuals school and family (Sawchuk & Buchhalter, 2015). Signposting was provided in some interventions (n=6) for participants and five interventions included a session with a review of the content covered.

Table 3: Intervention Components

References	Safety information	Explaining NES, mind-body connection and symptoms	Explaining different names for NES	Common predisposing factors	Conveying diagnosis as good news	Emphasis on symptoms being real and not fake/ the person is not "crazy"	Instilling hope that seizures may resolve over time	Describing personal experiences of overcoming seizures	Addressing stigma/ shame	Signposting	Understanding emotions	Influence of trauma and abuse	Influence of anger	Depression and anxiety	Identifying and challenging unhelpful thoughts	Trigger discrimination	QOL/ values	Problem solving skills	Taking responsibility for actions to manage seizures	Relaxation skills	Coping with stress	Avoidance behaviours	Group review session
(Zaroff et al., 2004)		X				X						X	X	X		X	X			X	X		X
(Wiseman et al., 2016)		X				X								X		X	X			X	X	X	
(Thompson et al., 2005)		X			X	X	X			X													
(Thompson et al., 2013)		X			X	X	X		X	X							X						
(Sawchuk & Buchhalter, 2015)	X	X																		X	X		
(Sarudiansky et al., 2020)		X				X					X					X				X			X
(Mayor et al., 2013)		X														X				X	X	X	X
(Libbon et al., 2019)		X				X			X							X			X	X	X		X
(Flewelling et al., 2020a)		X								X							X		X				
(Cope et al., 2017)		X													X					X		X	
(Cope et al., 2021)		X	X	X				X		X													
(Chen et al., 2014)	X	X			X											X		X	X	X	X		
(Baxter et al., 2012)		X		X			X			X						X		X	X	X		X	X
(Arain et al., 2007)		X																					
(Farias et al., 2003)		X		X	X	X	X			X		X				X			X	X			

(Vojvodic et al., 2006)	X		X												
(Gurcan et al., 2022)	X	X		Prior to group	Prior to group	Prior to group		X			X	X		X	X

Evaluations of interventions

Of the records included (n=17), 16 had some form of quantitative evaluation and two involved a qualitative aspect.

Quantitative Evaluations

Three interventions had a control group, though the majority of interventions (n=13) used a within groups pre-post analysis.

Seizure Reduction

Thirteen of the studies used reduction in seizure frequency/ cessation as a measure of evaluation. Four studies reported a significant reduction in the seizures: Flewelling et al (2020a), p=0.02; Cope et al (2017), p=0.032; Farias et al (2003), p=0.0001 and Vojvodic et al (2006), $p<0.05$. Three studies tested for and did not find a significant seizure reduction (Gurcan et al., 2022; N. Thompson et al., 2013; Wiseman et al., 2016). Six studies reported a reduction in seizures, but did not test for statistical significance and provided descriptive data on reductions in seizures ranging from reductions in 28.7% of participants (Zaroff et al., 2004) to 50% (Thompson et al., 2005). Of the 13, three studies (Mayor et al., 2013; Sarudiansky et al., 2020; Zaroff et al., 2004) reported that whilst some participants reported decreases in their seizures, other participants experienced increases in their seizures, ranging from 7.7% to 41.6% of participants reporting an increase.

Understanding and Acceptance of NES Diagnosis

Six studies explored participants understanding and acceptance of their diagnosis following the intervention, four of which used the Brief Illness Perception Questionnaire (BIPQ: Broadbent et al., 2006) to do so, and all found changes to be significant: Wiseman et al (2016) $Z = -3.027$, $p = 0.002$; Sarudiansky et al (2020), Z= -2.355, p=.019; Cope et al., (2017), p=< 0.001; Gurcan et al., (2022), p=<0.001. Another study (Cope et al., 2021) used

an idiosyncratic measure and found significant improvements for participants reporting on their experiences of their understanding of their diagnosis ($p<0.001$, $d=1.0$), beliefs about treatability ($p<0.001$, $d=0.55$), hope for recovery ($p<0.001$, $d=0.17$) and agreement with diagnosis ($p<0.001$, $d=0.33$). Furthermore another idiosyncratic measure of diagnosis acceptance found 66% (n=19) of participants accepted their diagnosis following the intervention, though there was not comparison data available for this prior to the intervention (Sawchuk & Buchhalter, 2015).

QOL

There was little consistency in terms of psychometric measures used to assess QOL factors in the eight studies examining it. The most frequently used measure (n=4) was the Work and Social Adjustment Scale (Mundt et al., 2002). Two studies (N. Thompson et al., 2013; Zaroff et al., 2004) used the Quality of Life in Epilepsy-31 (Vickrey et al., 1993), though results were not significant. No other studies used the same measures, making comparison of studies difficult. Seven studies measured statistical significance of QOL changes, of which four were statistically significant (Chen et al., 2014; Flewelling et al., 2020a; Gurcan et al., 2022; Wiseman et al., 2016), however all studies with significant findings used different QOL measures. One study (Mayor et al., 2013) reported on QOL but did not conduct measures of statistical significance.

Mental Health

Four studies examined mental health symptoms following the intervention including exploring emotional impact (Cope et al., 2017), coping skills, trauma symptoms, dissociation (Zaroff et al., 2004), psychological distress depression and anxiety (Cope et al., 2017; Flewelling et al., 2020a; Sarudiansky et al., 2020). All studies used measures of statistical significance to evaluate change, with significant improvements being found in psychological distress (Cope et al., 2017), self-reported and parent reported depression

scores (Flewelling et al., 2020a; Sarudiansky et al., 2020), state anxiety (Sarudiansky et al., 2020), coping skills and trauma symptoms (Zaroff et al., 2004). No significant difference was found in domains of anxiety across three studies (Cope et al., 2017; Flewelling et al., 2020a; Sarudiansky et al., 2020) and one study also found no differences in self-reported depression scores (Flewelling et al., 2020a)

Service Utilisation

Idiosyncratic measures of service utilisation were used in six studies and considered general functioning such as attending appointments, school and reduction in emergency department use. Four studies reported a reduction in emergency department use following intervention, with two noting the reduction in visits reached statistical significance (Flewelling et al., 2020a: $z = -2.68$, $p = 0.004$; Gurcan et al., 2022: $p = 0.04$) as well as a significant reduction in school absence (Flewelling et al., 2020a: $p = 0.02$). One study reported that 100% of participants made and kept an appointment with a mental health professional after the intervention (Thompson et al., 2013) and another reported that following the intervention, there was an increase in participants seeking psychological treatment (Mayor et al., 2013).

Qualitative Evaluations

The two studies involving qualitative components (Baxter et al., 2012; Sarudiansky et al., 2020) conducted thematic analyses of responses to interview data or questionnaire responses, the results can be seen in a narrative summary in Table 4. Gaining knowledge and finding answers to questions about NES was a common theme to both studies, though in interviews (Baxter et al., 2012), participants expressed concerns over the validity of the diagnosis for them given the efficacy of medication and feeling that the emotional explanation for the diagnosis was not sufficient for their experiences.

Table 4: Narrative summary of qualitative results

References	Data collection and analysis	Qualitative results
(Sarudiansky et al., 2020)	Descriptive thematic analysis of qualitative data gathered as part of the Pre-Post Psychoeducational Intervention Questionnaire	Concern and management of NES: • Prior to treatment, 41.67% of participants expressed concern over how people would respond to seeing their NES happen in responses to open ended questions on a pre-post questionnaire, following the intervention, no one reported concerns anymore. • After the workshop, 50% reported using skills that they had been taught in response to open-ended questionnaire questions. NES attributions: • Most participants spoke of psychosocial causes of NES prior to the workshop. Afterwards, one person changed their attributions from biological to psychosocial. Workshop experience: • All participants (n=12) said the group was a positive experience. • Most useful components were identified as: sharing the experience (n=6), learning coping strategies (n=5) and gaining knowledge about NES (n=2).
(Baxter et al., 2012)	Semi-structured interviews, analysed through thematic	Getting answers: • A general sense of participants needing information and the intervention allowing them to ask questions.

analysis

Understanding the link with emotion:

- Psychological explanations were challenging for participants, though 50% (n=6) noted that they had gained an understanding of the link between their emotions and seizures from the intervention.

Seeking a physiological explanation:

- 41.67% (n=5) of participants did not feel the emotional explanation for their seizures was sufficient and were either unable or not willing to notice possible emotional triggers.

Doubting the diagnosis:

- For 50% (n=6), the intervention had not provided a total explanation about the onset and continuation of their symptoms, with some people expressing concerns that "something was being missed".

The role of medication:

- Difficulties in fully understanding and "trusting" the NES diagnosis was identified as possibly linked for 33.33% (n=4) of participants due to a perceived impact of medication seeming to reduce their seizures

Finding a way forward:

- 25% (n=3) said that the intervention had given them a more positive perspective on the future.
- Of these participants, one described their symptoms as improving, and two said there was no change.
- A hope of resolving unanswered questions about causation was expressed by these participants

Discussion

Summary

This review systematically searched the evidence to ascertain the content and efficacy of psychoeducational interventions for reducing NES and increasing wellbeing. 17 studies were included, two of which had qualitative evaluations in their design.

Components of interventions

Interventions varied in what they offered as part of their psychoeducation and the time dedicated to them, though the majority were in a group format (n=10) and all included a general discussion of NES and the mind-body link, with seven interventions also stressing that symptoms were real and the patient was not "crazy". Given that research (Howlett et al., 2007; Thompson et al., 2009) suggests that failure to provide such information in a way that feels acceptable to the patient, results in mistrust and disengagement from support services, inclusion of these components in a sensitive way is crucial. Interventions also included components of instilling hope and portraying an NES diagnosis as good news, despite some research suggesting this is not always well received (McWilliams et al., 2016). Many interventions also entailed skill-based elements such as trigger discrimination, problem solving, coping with stress, relaxation skills and overcoming avoidance. Such skill-based elements are part of CBT protocols as well as being transdiagnostic to a range of interventions for other mental health concerns, such as anxiety and depression, that have high comorbidity in NES (Flewelling et al., 2020a; Karakis et al., 2014; Robson et al., 2018). Given that research also suggests that people with NES experience lower QOL and functioning (Reuber, 2008), it was encouraging to see interventions also covered broader considerations with four interventions covering QOL and values as well as two covering depression and anxiety, two covering shame and

stigma, two covering trauma and another focussing on emotions generally. However, despite interventions covering a wide range of relevant topics, and the majority reported that they were manualised (n=12), no two interventions were the same in their components and varied between single-sessions and up to ten weeks in length, demonstrating a lack of consistency in what is provided. Such findings highlight the need for replication of specific interventions to add validity to the evidence base and inform staffing levels.

Efficacy of interventions

Seizure frequency and understanding of diagnosis

Psychoeducational interventions that have been evaluated in their own right are scant in the literature and none identified by this review were randomised control trials. Outcome of the interventions varied across the identified studies. Of the 13 studies reporting data on seizure frequency, 12 reported reductions in seizures, however, only seven of these measured the statistical significance of the changes, with four studies reaching significance. Studies using the BIPQ (Broadbent et al., 2006) all found significant improvements in participants understanding of NES, and improvements were also found by studies using idiosyncratic or qualitative measures, though only two studies reported statistical significance and qualitative studies also highlighted conflict in how participants felt about the information provided and their personal experiences. Such improvements in understanding suggest interventions were able to convey sensitive information that was well-received, which is important in increasing engagement with future support (Howlett et al., 2007; Thompson et al., 2009).

QOL

Interventions measuring QOL were varied in the measures used making direct comparison of results difficult. However, of the eight studies, seven reported significant improvements

in QOL, suggesting that psychoeducation for NES can have implications for wider social functioning and perceptions of disability. However, due to limited data available and the studies included being varied in method, duration, format and content; it is not possible to conclude a direct causal effect from the current literature.

Wellbeing and service utilisation

Some studies utilised engagement with support as an outcome measure and a significant reduction in emergency department used was reported by two studies. Another reported that all participants made and attended a mental health appointment following the intervention (Thompson et al., 2013) and another found an increase in people seeking psychological treatment (Mayor et al., 2012). Such results are important as previous research has found that increased attendance at mental health appointments for those with NES is associated with increased QOL (Gagny et al., 2021). Increased attendance at appointments is also important as we have increasing evidence that psychological interventions for NES work (Goldstein et al., 2010) and the impact of psychoeducation as a standalone treatment is mixed and limited in its generalisability. Measures of assessment for mental wellbeing were also not consistent in application or results across studies, highlighting that psychoeducation as an intervention for NES needs consistency in its delivery and evaluation for more robust studies to be conducted and to be able to draw clear statistical conclusions.

Strengths and Limitations

This review has allowed for a comprehensive snapshot of the current evidence base for psychoeducation as an intervention for NES and has drawn attention to issues such as lack of replication of manualised interventions, small sample sizes and inconsistencies in outcome measures that currently make drawing conclusions difficult. Since this review took a broad scope to allow for an overview of the theoretical and methodological diversity

in the literature, the review includes a range of psychoeducational interventions for people with NES that was not limited to age or modality, and the variability of what different interventions provide has been highlighted. In future reviews such complexities in the varied scope of information may be controlled for by limiting inclusion criteria to a more specific definition of 'psychoeducation'; for example, classifying it as a didactic intervention occurring outside of the initial diagnostic session for NES and delivered by a healthcare professional for a pre-determined amount of time such as up to five sessions. However, due to large variability in the literature it may be that few studies would meet inclusion for such a study and so this would also need taking into account. A limitation of the review is that the studies included did not undergo quality assessment and were not examined for methodological rigor. Due to this, conclusions drawn lack context about the risk of bias in each study included and make it difficult to draw generalizable conclusions to the wider NES population and clinical practice. However, this did allow for the inclusion of grey literature that would not have otherwise been possible to include. A further limitation of this study is that it only included papers that were written in English, meaning that it is possible that eligible reports were missed. Furthermore, information about the components of each intervention (Table 3) were only described if they were explicit in the report of each study, thus some studies may have had components omitted due to lack of clarity.

Conclusions

There are a wide variety of approaches to providing psychoeducation for NES, all of which include crucial components of describing symptomology and the mind-body link; however no intervention was identical in its approach. Inconsistencies in reporting of outcomes and small sample sizes make it difficult to ascertain a direct impact of psychoeducation, though there is emerging evidence of seizure reduction and improved quality of life following at

least some of the interventions. Given the potential for long waiting lists for therapy in health settings, psychoeducation is likely to be a useful first line of treatment for NES for achieving remission in some clients and introducing psychological concepts to clients who then require further more intensive intervention. Furthermore, studies with child populations tended to consist of single-session interventions (n=3/4) with comparable results to longer interventions aimed at adults as well as the results between group and individual sessions being comparable, suggesting that interventions for adults could be adapted to be more efficient to meet service pressures.

Future Directions

Future research needs to replicate interventions on a larger scale to create robust data and allow for conclusions to be drawn about the validity of interventions that is not restricted by underpowered samples or variability of intervention content. The evidence base would also benefit from research being conducted to explore what the most effective components of psychoeducation for NES are for improving symptoms and QOL. Further exploration into the critical components of successful psychoeducation is required to determine if specific populations, comorbidities or frequency of NES mediates outcomes. As results between groups and individual sessions as well as longer and shorter interventions appear comparable, more research into a single-session group modality would be valuable to inform clinical practice.

References

Arain, A. M., Hamadani, A. M., Islam, S., & Abou-Khalil, B. W. (2007). Predictors of early seizure remission after diagnosis of psychogenic nonepileptic seizures. *Epilepsy & Behavior*, *11*(3), 409–412. https://doi.org/10.1016/j.yebeh.2007.07.017

Arksey, H., & O'Malley, L. (2005). Scoping studies: Towards a methodological framework. *International Journal of Social Research Methodology*, *8*(1), 19–32. https://doi.org/10.1080/1364557032000119616

Asadi-Pooya, A. A., & Sperling, M. R. (2015). Epidemiology of psychogenic nonepileptic seizures. *Epilepsy & Behavior*, *46*, 60–65. https://doi.org/10.1016/j.yebeh.2015.03.015

Baxter, S., Mayor, R., Baird, W., Brown, R., Cock, H., Howlett, S., House, A., Messina, J., Smith, P., & Reuber, M. (2012). Understanding patient perceptions following a psycho-educational intervention for psychogenic non-epileptic seizures. *Epilepsy & Behavior*, *23*(4), 487–493. https://doi.org/10.1016/j.yebeh.2011.12.002

Bowman, E. S. (1993). Etiology and Clinical Course of Pseudoseizures. *Psychosomatics*, *34*(4), 333–342. https://doi.org/10.1016/S0033-3182(93)71867-8

Broadbent, E., Petrie, K. J., Main, J., & Weinman, J. (2006). The Brief Illness Perception Questionnaire. *Journal of Psychosomatic Research*, *60*(6), 631–637. https://doi.org/10.1016/j.jpsychores.2005.10.020

Chen, D. K., Maheshwari, A., Franks, R., Trolley, G. C., Robinson, J. S., & Hrachovy, R. A. (2014). Brief group psychoeducation for psychogenic nonepileptic seizures: A neurologist-initiated program in an epilepsy center. *Epilepsia*, *55*(1), 156–166. https://doi.org/10.1111/epi.12481

Cohen, J. (1960). A Coefficient of Agreement for Nominal Scales. *Educational and Psychological Measurement*, *20*, 37–46.

Cope, S. R., Smith, J. G., Edwards, M. J., Holt, K., & Agrawal, N. (2021). Enhancing the communication of functional neurological disorder diagnosis: A multidisciplinary education session. *European Journal of Neurology*, *28*(1), 40–47. https://doi.org/10.1111/ene.14525

Cope, S. R., Smith, J. G., King, T., & Agrawal, N. (2017). Evaluation of a pilot innovative cognitive-behavioral therapy-based psychoeducation group treatment for functional non-epileptic attacks. *Epilepsy & Behavior*, *70*, 238–244. https://doi.org/10.1016/j.yebeh.2017.02.014

Farias, S. T., Thieman, C., & Alsaadi, T. M. (2003). Psychogenic nonepileptic seizures: Acute change in event frequency after presentation of the diagnosis. *Epilepsy & Behavior*, *4*(4), 424–429. https://doi.org/10.1016/S1525-5050(03)00143-4

Flewelling, K. D., Koehler, A., Shaffer, J., & Dill, E. J. (2020a). Medical and psychosocial outcomes of youth with psychogenic nonepileptic seizures: An observational study. *Epilepsy & Behavior*, *112*, 107383. https://doi.org/10.1016/j.yebeh.2020.107383

Flewelling, K. D., Koehler, A., Shaffer, J., & Dill, E. J. (2020b). Correlates of health-related quality of life in youth with psychogenic non-epileptic seizures. *Seizure*, *83*, 203–207. https://doi.org/10.1016/j.seizure.2020.09.030

Gagny, M., Grenevald, L., El-Hage, W., Chrusciel, J., Sanchez, S., Schwan, R., Klemina, I., Biberon, J., de Toffol, B., Thiriaux, A., Visseaux, J. F., Martin, M. L., Meyer, M., Maillard, L., & Hingray, C. (2021). Explanatory factors of quality of life in psychogenic non-epileptic seizure. *Seizure*, *84*, 6–13. https://doi.org/10.1016/j.seizure.2020.10.028

Goldstein, L. H., Chalder, T., Chigwedere, C., Khondoker, M. R., Moriarty, J., Toone, B. K., & Mellers, J. D. C. (2010). Cognitive-behavioral therapy for psychogenic nonepileptic seizures: A pilot RCT. *Neurology*, *74*(24), 1986–1994. https://doi.org/10.1212/WNL.0b013e3181e39658

Gurcan, L., McAllister, E., Gilmour, J., Green, D. J., McFarlane, F. A., Hadji-Michael, M., Heyman, I., & Stark, D. (2022). Improved understanding of non-epileptic seizures and reduced emergency health care usage following a single psychoeducational group for children and their parents. *Seizure*, *101*, 1–7. https://doi.org/10.1016/j.seizure.2022.07.001

Howlett, S., Grünewald, R. A., Khan, A., & Reuber, M. (2007). Engagement in psychological treatment for functional neurological symptoms—Barriers and solutions. *Psychotherapy: Theory, Research, Practice, Training*, *44*(3), 354–360. https://doi.org/10.1037/0033-3204.44.3.354

Karakis, I., Montouris, G. D., Piperidou, C., Luciano, M. S., Meador, K. J., & Cole, A. J. (2014). Patient and caregiver quality of life in psychogenic non-epileptic seizures compared to epileptic seizures. *Seizure*, *23*(1), 47–54. https://doi.org/10.1016/j.seizure.2013.09.011

Kozlowska, K., Chudleigh, C., Cruz, C., Lim, M., McClure, G., Savage, B., Shah, U., Cook, A., Scher, S., Carrive, P., & Gill, D. (2018). Psychogenic non-epileptic seizures in children and adolescents: Part II – explanations to families, treatment, and group outcomes. *Clinical Child Psychology and Psychiatry*, *23*(1), 160–176. https://doi.org/10.1177/1359104517730116

Libbon, R., Gadbaw, J., Watson, M., Rothberg, B., Sillau, S., Heru, A., & Strom, L. (2019). The feasibility of a multidisciplinary group therapy clinic for the treatment

of nonepileptic seizures. *Epilepsy & Behavior*, *98*, 117–123. https://doi.org/10.1016/j.yebeh.2019.06.032

Mayor, R., Brown, R. J., Cock, H., House, A., Howlett, S., Singhal, S., Smith, P., & Reuber, M. (2012). Short-term outcome of psychogenic non-epileptic seizures after communication of the diagnosis. *Epilepsy & Behavior*, *25*(4), 676–681. https://doi.org/10.1016/j.yebeh.2012.09.033

Mayor, R., Brown, R. J., Cock, H., House, A., Howlett, S., Smith, P., & Reuber, M. (2013). A feasibility study of a brief psycho-educational intervention for psychogenic nonepileptic seizures. *Seizure*, *22*(9), 760–765. https://doi.org/10.1016/j.seizure.2013.06.008

McWilliams, A., Reilly, C., McFarlane, F. A., Booker, E., & Heyman, I. (2016). Nonepileptic seizures in the pediatric population: A qualitative study of patient and family experiences. *Epilepsy & Behavior*, *59*, 128–136. https://doi.org/10.1016/j.yebeh.2016.03.029

Mellers, J. D. C. (2005). The approach to patients with "non-epileptic seizures". *Postgraduate Medical Journal*, *81*(958), 498–504. https://doi.org/10.1136/pgmj.2004.029785

Mundt, J. C., Marks, I. M., Shear, M. K., & Greist, J. M. (2002). The Work and Social Adjustment Scale: A simple measure of impairment in functioning. *British Journal of Psychiatry*, *180*(5), 461–464. https://doi.org/10.1192/bjp.180.5.461

Novakova, B., Harris, P. R., Rawlings, G. H., & Reuber, M. (2019). Coping with stress: A pilot study of a self-help stress management intervention for patients with epileptic or psychogenic nonepileptic seizures. *Epilepsy & Behavior*, *94*, 169–177. https://doi.org/10.1016/j.yebeh.2019.03.002

Operto, F. F., Coppola, G., Mazza, R., Pastorino, G. M. G., Campanozzi, S., Margari, L., Roccella, M., Marotta, R., & Carotenuto, M. (2019). Psychogenic nonepileptic seizures in pediatric population: A review. *Brain and Behavior*, *9*(12), e01406. https://doi.org/10.1002/brb3.1406

Ouzzani, M., Hammady, H., Fedorowicz, Z., & Elmagarmid, A. (2016). Rayyan—A web and mobile app for systematic reviews. *Systematic Reviews*, *5*(1), 210. https://doi.org/10.1186/s13643-016-0384-4

Page, M. J., McKenzie, J. E., Bossuyt, P. M., Boutron, I., Hoffmann, T. C., Mulrow, C. D., Shamseer, L., Tetzlaff, J. M., Akl, E. A., Brennan, S. E., Chou, R., Glanville, J., Grimshaw, J. M., Hróbjartsson, A., Lalu, M. M., Li, T., Loder, E. W., Mayo-Wilson, E., McDonald, S., … Moher, D. (2021). The PRISMA 2020 statement: An updated guideline for reporting systematic reviews. *BMJ*, *372*, n71. https://doi.org/10.1136/bmj.n71

Reuber, M. (2008). Psychogenic nonepileptic seizures: Answers and questions. *Epilepsy & Behavior : E&B*, *12*, 622–635. https://doi.org/10.1016/j.yebeh.2007.11.006

Robson, C., Myers, L., Pretorius, C., Lian, O. S., & Reuber, M. (2018). Health related quality of life of people with non-epileptic seizures: The role of socio-demographic characteristics and stigma. *Seizure*, *55*, 93–99. https://doi.org/10.1016/j.seizure.2018.01.001

Sarudiansky, M., Korman, G. P., Lanzillotti, A. I., Pico, M. M. A., Tenreyro, C., Paolasini, G. V., Wolfzun, C., Kochen, S., D'Alessio, L., & Myers, L. (2020). Report on a psychoeducational intervention for psychogenic non-epileptic seizures in Argentina. *Seizure - European Journal of Epilepsy*, *80*, 270–277. https://doi.org/10.1016/j.seizure.2020.04.008

Sawchuk, T., & Buchhalter, J. (2015). Psychogenic nonepileptic seizures in children—Psychological presentation, treatment, and short-term outcomes. *Epilepsy & Behavior*, *52*, 49–56. https://doi.org/10.1016/j.yebeh.2015.08.032

Su, M. T., McFarlane, F., Cavanna, A. E., Termine, C., Murray, I., Heidemeyer, L., Heyman, I., & Murphy, T. (2017). The English Version of the Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents (C&A-GTS-QOL): A Validation Study in the United Kingdom. *Journal of Child Neurology*, *32*(1), 76–83. https://doi.org/10.1177/0883073816670083

Thompson, N. C., Osorio, I., & Hunter, E. E. (2005). Nonepileptic Seizures: Reframing the Diagnosis. *Perspectives in Psychiatric Care*, *41*(2), 71–78. https://doi.org/10.1111/j.1744-6163.2005.00023.x

Thompson, N., Connelly, L., Peltzer, J., Nowack, W. J., Hamera, E., & Hunter, E. E. (2013). Psychogenic Nonepileptic Seizures: A Pilot Study of a Brief Educational Intervention: Psychogenic Nonepileptic Seizures: A Pilot Study of a Brief Educational Intervention. *Perspectives in Psychiatric Care*, *49*(2), 78–83. https://doi.org/10.1111/j.1744-6163.2012.00353.x

Thompson, R., Isaac, C. L., Rowse, G., Tooth, C. L., & Reuber, M. (2009). What is it like to receive a diagnosis of nonepileptic seizures? *Epilepsy & Behavior*, *14*(3), 508–515. https://doi.org/10.1016/j.yebeh.2008.12.014

Vickrey, B., Perrine, K. R., Hays, R. D., Hermann, B., Cramer, J., Gordon, J., Meador, K. J., & Devinsky, O. (1993). *Scoring manual for the quality of life in epilepsy inventory-89 (QOLIE-89)*. Santa Monica, CA: Rand. https://www.rand.org/content/dam/rand/www/external/health/surveys_tools/qolie/qolie31_scoring.pdf

Vojvodic, N., Ristic, A., Sokic, D., Jankovic, S., & Sindjelic, A. (2006). Presenting the definitive diagnosis of psychogenic nonepileptic seizures may have therapeutic effects. *Poster Session 1: Alternative Therapies*, *47*, 68–69. https://doi.org/10.1111/j.1528-1167.2006.00715_23.x

Whitehead, K., & Reuber, M. (2012). Illness perceptions of neurologists and psychiatrists in relation to epilepsy and nonepileptic attack disorder. *Seizure*, *21*(2), 104–109. https://doi.org/10.1016/j.seizure.2011.09.012

Wiseman, H., Mousa, S., Howlett, S., & Reuber, M. (2016). A multicenter evaluation of a brief manualized psychoeducation intervention for psychogenic nonepileptic seizures delivered by health professionals with limited experience in psychological treatment. *Epilepsy & Behavior*, *63*, 50–56. https://doi.org/10.1016/j.yebeh.2016.07.033

Zaroff, M., Charles, Myers, L., B. Barr, W., Luciano, D., & Devinsky, O. (2004). Group psychoeducation as treatment for psychological nonepileptic seizures. *Epilepsy & Behavior*, *5*(4), 587–592. https://doi.org/10.1016/j.yebeh.2004.03.005

Service Improvement Project: Decision making in NF2 Hearing Interventions

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Target journal: Psychology, Health and Medicine

This journal has been chosen due to the wide ranging scope of the topics it covers and the topic of hearing interventions for neurofibromatosis type two being relevant across all aspects of the journal. This journal also published a similar style study recently about decision making for interventions for single-sided deafness and so it is likely that this would complement that study.

Abstract

The project aimed to understand experiences of people with Neurofibromatosis type 2 (NF2) under the Oxford and South West service, in terms of psychological impact of NF2 and support required for decisions to take up hearing interventions (hearing aids, auditory brain stem implants (ABI) and cochlear implants (CI)). Fifty surveys were completed and analysed. Qualitative findings demonstrated physical symptoms contributed to emotional distress, social isolation and challenged sense of identity. Quantitative findings showed people generally felt informed and ready to take up hearing interventions, with many feeling satisfied with the outcome. People without CI/ABI expressed that surgery risk and visibility of devices were barriers. Service recommendations include increasing accessibility of psychological support before interventions, greater provision of information, and consideration of communication and emotional needs of patients.

Key words: Neurofibromatosis, NF2, Hearing, Audiology, Cochlear implant, Hearing aid, Auditory brain stem implant, Neurology, Oncology, Psychology, Adjustment.

Introduction

Outline of NF2 and treatments

Neurofibromatosis type 2 (NF2) is a genetic condition causing benign tumour growth on nerve cells leading to several debilitating symptoms. Symptoms include hearing loss, balance issues and tinnitus as the vestibular system is affected by tumours (Nerve Tumours UK, 2022a; NHS, 2021). NF2 is a rare condition with prevalence estimated to be 1/30,000 people (Nerve Tumours UK, 2022b). Treatments include medication, surgery and radiotherapy to reduce tumour growth and preserve hearing (Goutagny & Kalamarides, 2018; NHS, 2021; Phi et al., 2009). Very often patients are offered hearing aids and, in some circumstances, cochlear implants (CI) and auditory brain stem implants (ABI).

CI/ABI are surgical treatments involving implantation of a device within the skull, aimed at improving access to sound. The process of disease progression in NF2 is unpredictable with periods of stability and sudden aggravation/loss creating repeated setbacks for patients and need for further multidisciplinary support (Carillo et al., 2018).

Impact of NF2 on psychological wellbeing

Studies of patient wellbeing have found NF2 patients are negatively impacted in their psychological wellbeing and psychosocial functioning (Quarmby et al., 2019) with higher rates of depression, anxiety and perceived stress as well as lower self-esteem than the general population (Wang et al., 2012). Anxiety and depression scores were also higher than those found in other life-threatening medical conditions, e.g. cancer and coronary heart disease (Wang et al., 2012). Symptom onset typically occurs at around 20 years, with diagnosis occurring on average at 28 (Johns Hopkin Medicine, 2022); thus NF2 can impact early stages of adulthood, often altering people's expectations of life trajectory. Whilst Wang et al (2012) did not find a specific association between hearing loss and mental health difficulties, qualitative research has found hearing loss and facial palsy in NF2 populations led to social withdrawal, feelings of frustration, embarrassment and isolation (Patel et al., 2011). Neary et al., (2010) found 73% (n=62) rated hearing loss as their greatest problem and 65% cited hearing loss as the main causal factor in lower confidence. Additionally, 75% indicated hearing loss had a moderate or severe effect on their lives, with 43% reporting difficulties communicating with the most significant person in their life, and 78% reporting social communication difficulties. These findings highlight a role for psychological support.

Adapted models of psychosocial adjustment and coping offer explanations of how symptoms are experienced and what might impact optimal adjustment (e.g. (Kennedy & Duff, 2005)– see Appendix B). Research into other chronic conditions indicates mastery

over symptoms (e.g. pain management), absence of mental illness, and ability to perceive goals as obtainable indicate greater adjustment (Baum et al., 2012). Additionally, research demonstrates psychological interventions focusing on coping strategies improved outcomes in chronic conditions (McDonald et al.,2011; Turk et al., 2008). Despite this, there are no national standards or specifications for psychological input to patients with NF2.

Patient decision making regarding treatments

Complexity

Qualitative research into decision making for hearing interventions in non-NF2 populations shows people continually weigh up risks and benefits of interventions, guided by personal experiences and practical use (e.g. how comfortable the device was and how likely they were to need it professionally). Desire to regain hearing ability has also been influenced by a deficit-focussed medical approach (Underdown & Pryce, 2021). Barriers to proceeding with interventions included other health concerns, burden of multiple hospital visits, fear of stigma and not feeling fully informed about treatment. This highlights the need for patients to have appropriate information and time to discuss decisions about hearing interventions and likely benefits, as per the principles of the Collaborative Deliberation Model (Elwyn et al., 2014).

The decision making process for treatments in NF2 may be more complex, particularly for ABI/CI where outcomes may be less beneficial and less certain (Harris et al., 2017; Otto et al., 2002). This may leave patients feeling confused, despondent and less engaged.

Theoretical considerations

The NHS Long Term Plan emphasises the importance of personalised care and improved patient experiences, which is particularly relevant for decision making (NHS, 2022).

Decision making theories offer insight into factors impacting on patient appraisals and how clinicians can facilitate discussions, with consideration of cost-benefit analyses, more complex social influences, unconscious processes and instincts (Edwards & Elwyn, 2009). These principles are particularly relevant to healthcare where people may be biased by how information is presented to them, e.g. being influenced by their relationship with the clinician (Chaiken, 1980) or by how outcome statistics are presented to them (Cuite et al., 2008; Gigerenzer & Edwards, 2003). The Collaborative Deliberation Model (Elwyn et al., 2014) proposes that for effective decisions to be made which are personalised to the patient, five key steps must occur: constructive engagement; recognition of alternative actions, comparative learning, preference construction and elicitation, and finally, preference integration. Therefore, for effective healthcare decisions about hearing loss, patients must be actively engaged by clinicians to help them think through and make the choices for themselves. These principals have been used in the present study to inform questions about patient experience.

The Collaborative Deliberation model has been found to facilitate conversations about obesity and its physical health complications in ways that were acceptable to patients who reported feeling heard and motivated to engage in health decisions based on their priorities (Luig et al., 2019). Whilst the Collaborative Deliberation Model does not provide a framework ascertaining why people make decisions, it provides a framework for healthcare professionals to facilitate personalised care and exploration of complex health decisions.

Uptake of hearing interventions within the service

Economic theories of decision making would suggest high ABI/CI uptake in the NF2 population given the potential benefits to hearing. However, an audit of NF2 services across England revealed ABI/CI usage after implantation was lower than the general population, with 38% of implants not being used. Initial implantation rate at the Oxford

and South West NF2 Service (O&SW NF2 Service) was also 50% lower than other services. It is unclear whether differences in clinical treatment pathways or access to psychological support between centres plays a role in this lower uptake.

Given low usage rates post implantation and low uptake in the O&SW NF2 Service, this study aimed to understand more about patients' experiences of hearing interventions and support provided for these. As CI/ABI is offered in the context of a complex hearing intervention pathway, the study also aimed to explore patient experiences of hearing aids. Furthermore, given the lack of service specification for psychological input in NF2 services, the study aimed to explore the impact of NF2 on patients' wellbeing to make recommendations about how they might be supported.

Research questions

Key study questions were identified to drive service improvements in the area of hearing loss and hearing intervention support (Table 1).

Table 1: Key questions and how they will drive improvements

Question	**Potential area of improvement**
How do patients with NF2 experience NF2?	Identification of psychological support needs to inform treatments offered
How do patients with NF2 experience hearing interventions in terms of information provision, decision making and general support from the team?	Identification of what further help and information the service may need to offer to support the decision making process
What improvements would patients with NF2 like to see in terms of support with hearing loss and interventions?	Identification of patient-reported service needs

Methodological and ethical considerations

The project obtained audit approval from Oxford University Hospitals NHS Trust (Appendix C). A voluntary and anonymous survey was distributed via email and paper

copies. This method was chosen to reduce burden to patients and optimise confidentiality and accessibility. Adult patients diagnosed with NF2 who had received any input from the O&SW NF2 Service were included to capture broad feedback.

Methods

Survey design

Stakeholders within the service (team members and patients) and a service user charity were consulted as part of survey development to augment content validity. The survey was designed by the researchers with the key research questions in mind and piloted by a patient volunteer to ensure readability and to identify any issues with technology or question flow. The survey contained free text questions as well as fixed choice and Likert scale questions (Appendix D).

Inclusion and exclusion criteria

All patients who attended the O&SW NF2 Service since its commencement in 2010 are recorded on the database. Given the chronic nature of NF2 and the large catchment area the majority of patients remain with the service. The database was searched for all adult patients with a NF2 diagnosis. Two hundred and forty patients were identified and contacted by email or post and asked to complete a survey.

Those choosing to take part were screened through a question asking if they had a NF2 diagnosis in case of inaccuracies in the database or the link being shared inappropriately.

Analysis

Qualitative analysis

Free-text answers were analysed using inductive content analysis (Kyngäs, 2020) assisted by NVivo software to determine themes and codes (Appendix E and F). Data was

categorised into themes under the research questions and participant's words were used when possible (Robson, 2002). The number of times a theme was found in the data was recorded in order to quantify the relative contribution of the theme.

Coder 1 (author) undertook data familiarisation and coding, discussing this with coder 2 (service psychologist). The validity and reliability of themes were checked using an independent rater who was given 25% of the data to code. The independent rater had experience of qualitative analysis but was unfamiliar with NF2 and how codes had been developed to encourage data driven analysis. Inter-rater reliability was determined through percent agreement due to theme overlap meaning data was not suitable to report a Cohen's Kappa co-efficient (Gwet, 2014; McHugh, 2012). For data related to patient experiences of NF2, the inter-rater reliability was moderate (68% agreement) and upon discussion it was recognised that two codes could be collapsed into one as they were frequently coded for each other and accounted for much of the disagreement between raters. In data about experiences of hearing interventions and improvements, the inter-rater reliability was an acceptable level (83% and 81% respectively).

Quantitative analysis

Quantitative data was analysed through descriptive statistics using SPSS software.

Results

Initially 65 responses were recorded. Fourteen did not provide data beyond demographics and one said they did not know if they had a diagnosis of NF2 so were excluded from analysis, leaving a total of 50 participants. Five participants did not answer questions about hearing interventions or service improvement. Their responses were therefore analysed only in relation to how NF2 impacted them. Furthermore, boxes were missed in some paper copies of response forms, meaning some question response rates appear lower.

Demographics

Fifty-two percent of participants were male (n=26) and ages varied from 18-25 to 76+ years with each age decile being well represented (Appendix G).

Six participants (12%) out of 50 had a family member with NF2 and 16/50 (32%) had received support from another NF2 team. Sixteen out of forty-six (34%) said someone in their family had hearing difficulties, though this was not always from NF2. The highest reported symptom was hearing loss (Appendix G2), with 92% reporting it (n=46/50). Balance impairments were reported by 78% (n=39/50; Appendix G3) and 44% (n=22/50) reported impacts on mental health and self-esteem.

Participants were asked what hearing interventions (hearing aid, CI/ABI) they were using or had used previously (Table 2). Thirty-five (77.8%) had used hearing aids currently or historically, with 7.0% using CI (n=3) and 4.7% using ABI (n=2). Based on the team database for interventions, response rates for each subgroup were: hearing aids: 27%; CI: 33% and ABI: 22%. Approximately one in four people offered CI, and approximately one in three offered ABI took up the interventions.

Table 2: Uptake and offers of hearing interventions

Intervention	**Offered and took up (%)**	**Offered and declined (%)**	**Never offered (%)**
Hearing Aid (n=45)	77.8	8.9	13.3
Cochlear Implant (n=43)	7.0	23.3	69.8
Auditory Brain Stem Implant (n=43)	4.7	9.3	86.0

N= number of participants completing this section of the survey

How do patients with NF2 experience it?

Qualitative data highlighted the burden and impact of NF2 with eight themes relating to wellbeing identified (Table 3).

Table 3: Themes relating to impact of NF2 on psychological wellbeing

Theme	**Incidence in data (n)**
Burden of physical symptoms: "What a cruel trick our bodies play on us"	54
Social impact and withdrawal: "I often feel left out"	44
Emotional burden: "Thinking about it makes me feel bad"	42
Impact on flexibility and adaptation to cope: "I've been able to adapt, but certain scenarios are still difficult"	26
Impact on identity and confidence: "It feels like sometimes there are no parts of me which are not affected"	18
"Career progression has been significantly impacted"	9
Sense of being lucky with NF2 as a whole: "I am lucky in that my NF2 is both mild and mosaic"	8

N.B: Incidence of themes may be higher than respondent number due to content analysis procedures

Analysis suggested themes were highly linked, with quotes often fitting into more than one theme (Appendix G3 and G4), with physical symptoms leading to feelings of isolation from others and stress along with anxiety about pressure placed on loved ones and disruption to career goals. Such overlap can be exemplified by one person's response:

"NF2 has completely taken over my life and everything I had ever known". As seen above, themes covered a wide range and there was much overlap between themes. For example, themes associated with symptoms also interlinked with a sense of challenge to identity and confidence, with people finding they needed to adapt in numerous ways to cope and continue engaging with life: "I went to lip reading classes for three years and mostly use this to get by".

Hearing loss expectations

Just under 50% of participants (n=22; 48.9%) described their hearing loss as being in line with their expectations, with 64.4% (n=29) feeling they had been given enough information about hearing loss and 46.6% (n=21) feeling they had been given enough emotional support (Table 4).

Table 4: Expectations of hearing loss and support given (n=45)

	Strongly agree (%)	**Agree (%)**	**Neither agree nor disagree (%)**	**Disagree (%)**	**Strongly disagree (%)**	**Don't know (%)**
My experience of hearing loss is in line with what I expected to happen	15.6 (n=7)	33.3 (n=15)	22.2 (n=10)	11.1 (n=5)	8.9 (n=4)	8.9 (n=4)
I am satisfied with the amount of information I was given by the NF2 team about hearing loss	20.0 (n=9)	44.4 (n=20)	22.2 (n=10)	6.7 (n=3)	2.2 (n=1)	4.4 (n=2)
The information the NF2 team have given me about hearing loss was easy to understand	22.2 (n=10)	51.1 (n=23)	17.8 (n=8)	4.4 (n=2)	2.2 (n=1)	2.2 (n=1)
The information the NF2 team have given me about hearing loss was relevant and helpful to me	26.7 (n=12)	37.8 (n=17)	24.4 (n=11)	4.4 (n=2)	0.0 (n=0)	6.7 (n=3)
I was given enough emotional support to adjust to the impact of hearing loss	13.3 (n=6)	33.3 (n=15)	31.1 (n=14)	8.9 (n=4)	11.1 (n=5)	2.2 (n=1)

How do patients with NF2 experience hearing interventions in terms of information provision, decision making and general support from the team?

Information provision about hearing devices

Participants were asked about their satisfaction with the amount of information they were given regarding hearing devices, the support given and their satisfaction with the device (Table 5). Most people reported feeling supported and ready to have their hearing aids and most were satisfied with the outcome, though five reported it had not improved their communication and were not satisfied. All four participants with CI/ABI reported feeling committed to engaging in rehabilitation and felt supported in their decision. Most people were satisfied with the outcome, though one person reported it had not improved their communication and they were not satisfied.

Decision making processes of hearing interventions

The process of deciding to have a hearing aid or CI/ABI was explored (Table 6) as well as attitudes towards CI/ABI of four hearing aid participants (Table 7).

Table 5: People's experiences of having hearing aids (n=35) and CI/ABI (n=4) fitted reported in percentage

	Strongly Agree (%)		**Agree (%)**		**Neither agree nor disagree (%)**		**Disagree (%)**		**Strongly disagree (%)**		**Don't know (%)**	
	Hearing aid	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*
I was given enough information about the procedure of having my hearing device fitted.	40.0 (n=14)	25.0 (n=1)	40.0 (n=14)	75.0 (n=3)	17.1 (n=6)	0.0 (n=0)	2.9 (n=1)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)
I felt supported to make the decision to have this	40.0 (n=14)	25.0 (n=1)	37.1 (n=13)	75.0 (n=3)	14.3 (n=5)	0.0 (n=0)	8.6 (n=3)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)
I understood the importance of wearing my device and practicing listening with my device	42.9 (n=15)	50.0 (n=2)	40.0 (n=14)	25.0 (n=1)	8.6 (n=3)	0.0 (n=0)	8.6 (n=3)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	25.0 (n=1)
I felt ready and able to take part in the rehabilitation process (attend appointments, device adjustment, wearing the device in different settings)	29.4 (n=10)	50.0 (n=2)	32.4 (n=11)	50.0 (n=2)	26.5 (n=9)	0.0 (n=0)	11.8 (n=4)	0.0 (n=0)	0.0 (n=0)	25.0 (n=1)	0.0 (n=0)	0.0 (n=0)

My hearing device has helped me access sound and helped my communication	40.0 (n=14)	50.0 (n=2)	28.6 (n=10)	25.0 (n=1)	17.1 (n=6)	0.0 (n=0)	5.7 (n=2)	0.0 (n=0)	8.6 (n=3)	25.0 (n=1)	0.0 (n=0)	0.0 (n=0)
Overall I am satisfied with the outcome of my hearing device	31.4 (n=11)	50.0 (n=2)	40.0 (n=14)	25.0 (n=1)	11.4 (n=4)	0.0 (n=0)	8.6 (n=3)	0.0 (n=0)	8.6 (n=3)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)

Table 6: How people found the decision process of having a hearing aid (n=35) and CI/ABI (n=4)

	Strongly Agree (%)		**Agree (%)**		**Neither agree nor disagree (%)**		**Disagree (%)**		**Strongly disagree (%)**		**Don't know (%)**	
	Hearing aid	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*	*Hearing aid*	*CI/ABI*
I had time to think about the information I was given before making a decision	34.1 (n=11)	75.0 (n=3)	48.6 (n=17)	25.0 (n=1)	11.4 (n=4)	0.0 (n=0)	8.6 (n=3)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)
I made the decision based on scientific and medical information that I was given	20.0 (n=7)	75.0 (n=3)	54.3 (n=19)	25.0 (n=1)	17.1 (n=6)	0.0 (n=0)	8.6 (n=3)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)
I made the decision based on experiences of other people I know	5.7 (n=2)	0.0 (n=0)	20.0 (n=7)	25.0 (n=1)	20.0 (n=7)	0.0 (n=0)	37.1 (n=13)	25.0 (n=1)	14.3 (n=5)	50.0 (n=2)	2.9 (n=1)	0.0 (n=0)

I made the decision based on logistical factors such as family and ability to travel to appointments	14.3 (n=5)	0.0 (n=0)	28.6 (n=10)	25.0 (n=1)	20.0 (n=7)	0.0 (n=0)	28.6 (n=10)	25.0 (n=1)	8.6 (n=3)	50.0 (n=2)	0.0 (n=0)	0.0 (n=0)
I made the decision on gut instincts	14.3 (n=5)	0.0 (n=0)	31.4 (n=11)	0.0 (n=0)	20.0 (n=7)	75.0 (n=3)	20.0 (n=7)	25.0 (n=1)	14.3 (n=5)	0.0 (n=0)	0.0 (n=0)	0.0 (n=0)
I was concerned about people being able to see that I was wearing a hearing device	28.6 (n=10)	0.0 (n=0)	14.3 (n=5)	50.0 (n=2)	11.4 (n=4)	0.0 (n=0)	14.3 (n=5)	25.0 (n=1)	25.7 (n=9)	25.0 (n=1)	5.7 (n=2)	0.0 (n=0)
I was given information about the possible risks and benefits to aid my decision	14.3 (n=5)	25.0 (n=1)	28.6 (n=10)	50.0 (n=2)	25.7 (n=9)	25.0 (n=1)	25.7 (n=9)	0.0 (n=0)	2.9 (n=1)	0.0 (n=0)	2.9 (n=1)	0.0 (n=0)

Table 7: Perspectives on CI/ABI by hearing aid users

Theme	Incidence in data	Illustrative quotes
Perceptions of physical risk of CI/ABI	4	*"I'd be concerned that it might negatively affect the quality of my hearing. Also, the idea of surgery and something so permanent is off putting."*
		"Cochlear - declined as it did not convince me that it would work all time and did not want to have this in my head…it seemed uncomfortable."
		"Cochlear/brainstem implants have been offered but the decision has been postponed until the still usable hearing cannot be preserved anymore… success cannot be guaranteed and on the side where I still have a more or less working hearing (with hearing aid) is not yet worth to sacrifice."
		"I wouldn't have it as there is the possibility of losing my hearing as a result of having it put in.....no thank you"

Experiences of people following use of hearing devices

Qualitative comments revealed people's experiences of hearing aids meeting and not meeting expectations, concerns over how devices were viewed by others and practical frustrations they experienced with their device (Table 8).

Table 8: Themes relating to experiences of hearing aids

Theme	**Incidence in data (n)**
Meeting or exceeding expectations: “Hearing aids are the closest thing to a miracle that I can imagine”	37
Hearing aid as vital for functioning: “I would not have been able to function if I did not take it up.”	31
Not meeting expectations: “the sound quality was poor and music, especially, was so distorted I couldn't bear it”	26
Fear of drawing attention to difference: “I wanted to appear more normal”	11
Practical concerns: “Find it too bulky”	11
Social benefits of the hearing aid: “I feel a little less cut off from the world when I use them”	9
Trust in professional opinion: “I have never declined to try or consider positioned options”	9
Deterioration in hearing leading to ceasing use: “it didn’t work anymore”	4

People reported feeling a sense of trust in the professionals to advise them about the “best thing to help” with one person saying “if it is felt by the NF2 team that [a hearing aid] is required I recommend giving it a go”. Many reported feeling their hearing aid had met their expectations and had provided social benefits and was vital to their daily life (Appendix Table F5) as illustrated by one person commenting: “Since having it installed I have been able to hear much more of what people are saying”.

However, some found the hearing aid did not meet their expectations, and it created practical concerns about battery life and technological requirements. Others also worried about how it looked (Appendix Table F6). Some people also found their hearing had declined and had stopped using the hearing aid: “Hearing aid wasn't powerful enough as

hearing was getting worse". Data illustrated both the improvements and limitations of having the CI/ABI (Table 9).

Table 9: Themes and quotes associated with CI/ABI use

Theme	Incidence in data (n)	Illustrative Quotes
Successes of CI/ABI	5	*"With the cochlear implant switch on, I feel part of the world. Without it- isolation!"* *"I can hear a lot better and I can join in conversations"*
Limitations of CI/ABI	4	*"Initially the cochlear implant was worse than useless because the hearing on my other side was ok. Once the other side deteriorated, the cochlear implant had a huge impact, giving me consonants again. I could understand almost zero speech for a while, but using the cochlear implant in the right environments…I can understand almost everything"* *"I'm not 100% sure it was necessary for me, I think I could have had gamma ray knife and not have ended up quite so wonky."*

What improvements would patients with NF2 like to see in terms of support with hearing loss and interventions?

Participants were forthcoming with both compliments for the team and recommendations (Appendix H). Recommendations for improvements are shown in Table 10.

Table 10: Patient recommendations for service improvement

Theme	Subthemes	Incidence in data (n)
Recommendations for improvement	Scaffolding of where to find help and information	11
	Information about hearing technology	11
	Need for more follow ups	7
	Awareness of emotional impact	6
	Greater awareness of communication needs	5

Verbatim quotes regarding patient recommendations can be found in Appendix Table G7.

Discussion

This project sought to explore patient experiences of being offered hearing interventions with a broader aim of better understanding the impact of NF2 on wellbeing, in order to make recommendations for service improvement. Participants reported a range of impacts from NF2: the burden of physical symptoms having a knock-on effect on other areas of their life such as their emotions, identity and social lives, with the need to adapt to NF2 being instrumental for overcoming difficulties. Patients described how NF2 affected many aspects of their lives, which emerged in how themes overlapped and interconnected. Such overlap highlights a need for comprehensive and psychologically informed care due to the pervasive impact on wellbeing. This echoes research and models of NF2 suggesting that repeated adjustment is needed for optimal psychological health (Carillo et al., 2018; Patel et al., 2011; Quarmby et al., 2019). As hearing loss is often cited as having the largest impact in NF2, more multidisciplinary focus in this area may be needed to improve and concentrate support, taking into account the context that hearing loss brings to other disabilities and symptoms experienced in NF2. Given psychologists abilities to formulate complex presentations, and consider multiple domains of people's lives, they are well placed to lead in focussing on adjustment to hearing loss in NF2.

Collaborative decision making principles suggest decisions are based on evaluation of a likely outcome and the benefits of said outcome in relation to other personal and social factors (Elwyn et al., 2014). The data from this project suggested people with NF2 repeatedly consider many components of likely success, social implications and perceived dangers when making decisions about hearing interventions. Those who had surgical interventions shared that previous hearing aids were no longer "powerful enough" and going "fully deaf", created a need to seek benefits of other interventions. Others who had

less profound hearing difficulties were more concerned about the risks of surgical procedures and social implications of having a visible hearing device, potentially reflecting that perceived benefits of such devices are lesser at their current hearing abilities. The decision making literature also suggests people think they have made decisions based on logic, but such processes actually tend to be more instinctive or biased by their relationship with a healthcare provider (Chaiken, 1980). The data suggests that although people were offered CI/ABI procedures, some declined them despite expressing trust in the opinions of professionals in the team. Participants also said they felt they had enough information and had based their decision on scientific and medical information given to them. Such data therefore suggests that decisions for CI/ABI in NF2 may not be subject to relational and heuristic factors in the same way other health decisions are. It is possible that participants were offered, but not explicitly recommended these interventions by the team; however, this nuance was not captured in the data. Participants also responded with differing levels of agreement around whether gut instincts had influenced their decision; 42.9% (n=16) of people with hearing aids agreed they had responded to gut instincts in their decision to have a hearing aid, but there was no agreement in the CI/ABI group. Such results suggest invasiveness of treatment may mediate how people approach the decision making, particularly as non-CI/ABI users expressed concern over the risk of undergoing these procedures. This is in line with postulations that perceptions of risk and how risk information is presented impacts on treatment decisions (Cuite et al., 2008; Gigerenzer & Edwards, 2003) .

Most participants reported feeling well supported by the O&SW NF2 Service and trusted advice they provided. Participants also said they had enough time to think before making a decision about a hearing aid, indicating the importance of weighing up information and therapeutic relationships in long term condition services. There was variation in other

factors influencing decision making, such as others experiences and concern over the visibility of the device.

28.6% (n=10) of participants with hearing aids reported that they had not been given information about the risks and benefits of a hearing aid to help their decision, emphasising a need for greater emphasis on providing risk and benefit information, in line with patient centred care and collaborative decision making principles (Edwards & Elwyn, 2009).

Of the CI/ABI group 50% said people seeing them wear the device was a concern for them. When asked if they would recommend a CI/ABI to someone else with NF2, 50% said yes, but 50% were unsure.

Service implications

Based on the above results, six key recommendations are made for service improvements:

1. Offer a specific appointment to discuss beliefs and expectations about device outcome alongside existing provision of risks/benefits information. This may be best offered by the team psychologist due to being external to other medical processes and being able to use psychological principles to aid understanding and integration of information. An increase in psychological provision would likely need reviewing in terms of staffing to meet this need.
2. Consider involvement of patients in provision of information and support for people offered interventions (could be offered as a support group/online resources/quotes/stories of how something helped/tips for rehabilitation). This would aid in fostering psychologically-informed care and also allow patients to 'myth bust' together about devices as well as speaking to specialist clinicians.

3. Improve access to information about hearing device technology and consider what format to provide this (e.g. leaflet, signposting, noticeboard).
4. Provide more information and develop a clear referral/information pathway for lip reading as this may aid decisions around what patients try before invasive surgery
5. Consider use of pre-clinic appointment preference sheet to establish individualised communication needs in consultations.
6. Consider development of technology within waiting areas so patients are not reliant on purely auditory cues to know they have been called for their appointment.

These findings are due to be disseminated to patients in the service (Appendix I) as well as shared with other clinicians working within the NF2 field. It is hoped that these findings will encourage staff to be more psychologically informed in their care, as well as highlighting a need for the development of how communication aids are used in the service, inclusive of ways to present information about NF2 and relevant interventions to patients.

Study limitations

Despite the response rate being proportional to number of interventions given by the team across the groups, response rates were reasonably low compared to the number of people invited to take part. The total number of participants for CI/ABI interventions was four people, thus findings relating to CI/ABI are limited and it is unclear if those who did not respond opted out due to having bad experiences. Future research may want to consider how to get more feedback from this group, e.g. by asking for feedback after interventions or using a purposive sampling method. The overall sample was also heterogeneous as some participants may have only had contact with the team a couple of times many years ago and others may have had many years of involvement. This disparity could not be screened for due to the anonymous nature of the survey. Five participants did not answer questions

about hearing interventions or service development and some participants missed questions. However, given the rarity of NF2 and CI/ABI use, any information about people's experiences is still a valuable addition to the knowledge base.

At least 17.8% of people (n=8) chose 'neither agree nor disagree' with a statement about their experiences of hearing loss and expectations. This led to lack of clarity as to what may need to be improved. It may not be helpful to include these options in future projects seeking feedback.

Cohen's Kappa (McHugh, 2012) is considered to reduce the likelihood of agreement through chance between raters. However, this was not suitable for the data collected in the present study due to the codes overlapping between themes. Raters resolved any disagreement between codes and themes through discussion to identity the discrepancy in understanding and create a shared interpretation.

Future directions

This project provided insight into how people experience NF2, hearing interventions and support from the O&SW NF2 Service through the use of self-reported qualitative and quantitative questionnaire data. The project highlighted the pervasive nature of NF2 and viewpoints on different hearing interventions. Recommendations emphasised the need for psychologically informed care, adaptations for hearing loss in clinics and clear provision of information for patients. Future work would benefit from in depth interviews with patients who have taken up or declined CI/ABI to get richer feedback about their experiences. An audit of numbers of follow-up appointments offered following hearing interventions, their purpose (such as tuning hearing aid) and outcomes (e.g. successful or unsuccessful use of hearing device) would also provide valuable information and identify other barriers that patients may be facing.

References

Baum, A., Revenson, T. A., & Singer, J. (2012). *Handbook of Health Psychology: Second Edition*. Taylor & Francis.

Carillo, C., Kiening, M., Bergheimer, S., & Kalamarides, M. (2018). Psychological follow-up care of neurofibromatosis type 2 patients and their relatives. *Neurochirurgie*, *64*(5), 381–385. https://doi.org/10.1016/j.neuchi.2016.09.008

Chaiken, S. (1980). Heuristic Versus Systematic Information Processing and the Use of Source Versus Message Cues in Persuasion. *Journal of Personality and Social Psychology*, *39*(5), 752.

Cuite, C. L., Weinstein, N. D., Emmons, K., & Colditz, G. (2008). A Test of Numeric Formats for Communicating Risk Probabilities. *Medical Decision Making*, *28*(3), 377–384. https://doi.org/10.1177/0272989X08315246

Edwards, A., & Elwyn, G. (2009). *Shared Decision-making in Health Care: Achieving Evidence-based Patient Choice*. OUP Oxford.

Elwyn, G., Lloyd, A., May, C., van der Weijden, T., Stiggelbout, A., Edwards, A., Frosch, D. L., Rapley, T., Barr, P., Walsh, T., Grande, S. W., Montori, V., & Epstein, R. (2014). Collaborative deliberation: A model for patient care. *Patient Education and Counseling*, *97*(2), 158–164. https://doi.org/10.1016/j.pec.2014.07.027

Gigerenzer, G., & Edwards, A. (2003). Simple tools for understanding risks: From innumeracy to insight. *BMJ : British Medical Journal*, *327*(7417), 741–744.

Goutagny, S., & Kalamarides, M. (2018). Medical treatment in neurofibromatosis type 2. Review of the literature and presentation of clinical reports. *Neurochirurgie*, *64*(5), 370–374. https://doi.org/10.1016/j.neuchi.2016.09.004

Gwet, K. L. (2014). *Handbook of Inter-Rater Reliability, 4th Edition: The Definitive Guide to Measuring The Extent of Agreement Among Raters*. Advanced Analytics, LLC.

Harris, F., Tysome, J. R., Donnelly, N., Durie-Gair, J., Crundwell, G., Tam, Y. C., Knight, R. D., Vanat, Z. H., Folland, N., & Axon, P. (2017). Cochlear implants in the management of hearing loss in Neurofibromatosis Type 2. *Cochlear Implants International*, *18*(3), 171–179. https://doi.org/10.1080/14670100.2017.1300723

Johns Hopkin Medicine. (2022, January 4). *Neurofibromatosis 2*. Neurofibromatosis 2. https://www.hopkinsmedicine.org/neurology_neurosurgery/centers_clinics/pediatric-neurology/conditions/neurofibromatosis/nf2.html

Kennedy, P., & Duff, J. (2005). *Handbook of Clinical Health Psychology*. John Wiley & Sons.

Kyngäs, H. (2020). Inductive Content Analysis. *The Application of Content Analysis in Nursing Science Research*, 13–21. https://doi.org/10.1007/978-3-030-30199-6_2

Luig, T., Elwyn, G., Anderson, R., & Campbell-Scherer, D. L. (2019). Facing obesity: Adapting the collaborative deliberation model to deal with a complex long-term problem. *Patient Education and Counseling*, *102*(2), 291–300. https://doi.org/10.1016/j.pec.2018.09.021

McDonald, P. E., Zauszniewski, J. A., Bekhet, A. K., DeHelian, L., & Morris, D. L. (2011). The effect of acceptance training on psychological and physical health outcomes in elders with chronic conditions. *Journal of National Black Nurses' Association: JNBNA*, *22*(2), 11.

McHugh, M. L. (2012). Interrater reliability: The kappa statistic. *Biochemia Medica*, *22*(3), 276–282.

Neary, W. J., Hillier, V. F., Flute, T., Stephens, D., Ramsden, R. T., & Evans, D. G. R. (2010). Use of a closed set questionnaire to measure primary and secondary effects of neurofibromatosis type 2. *The Journal of Laryngology & Otology*, *124*(7), 720-728.

Nerve Tumours UK. (2022a, January 4). *How does NF2 affect you?* https://nervetumours.org.uk/what-are-nerve-tumours/what-is-nf2/how-does-nf2-affect-you

Nerve Tumours UK. (2022b, January 4). *What is NF2?* https://nervetumours.org.uk/what-are-nerve-tumours/what-is-nf2

NHS. (2021, April 27). *Neurofibromatosis type 2*. https://www.nhs.uk/conditions/neurofibromatosis-type-2/

NHS, N. L. T. (2022). *NHS Long term Plan*. NHS Long Term Plan. https://www.longtermplan.nhs.uk/online-version/overview-and-summary/

Otto, S. R., Brackmann, D. E., Hitselberger, W. E., Shannon, R. V., & Kuchta, J. (2002). Multichannel auditory brainstem implant: Update on performance in 61 patients. *Journal of Neurosurgery*, *96*(6), 1063–1071. https://doi.org/10.3171/jns.2002.96.6.1063

Patel, C. M., Ferner, R., & Grunfeld, E. A. (2011). A qualitative study of the impact of living with neurofibromatosis type 2. *Psychology, Health & Medicine*, *16*(1), 19–28. https://doi.org/10.1080/13548506.2010.516363

Phi, J. H., Kim, D. G., Chung, H.-T., Lee, J., Paek, S.-H., & Jung, H.-W. (2009). Radiosurgical treatment of vestibular schwannomas in patients with neurofibromatosis type 2. *Cancer*, *115*(2), 390–398.

Quarmby, L. M., Dalton, L. J., Woolrich, R. A., Clarke, R., Emmanouil, B., Patel, S., Rands, G., Parry, A., & Halliday, D. (2019). Screening and intervening: Psychological distress in neurofibromatosis type 2 (NF2). *Psycho-Oncology*, *28*(7), 1583–1587. https://doi.org/10.1002/pon.5099

Robson, C. (2002). *Real World Research* (2nd ed.). Blackwell Publishing.

Underdown, T., & Pryce, H. (2021). How do patients decide on interventions for single sided deafness? A qualitative investigation of patient views. *International Journal of Audiology*, *0*(0), 1–10. https://doi.org/10.1080/14992027.2021.1951853

Wang, D. L., Smith, K. B., Esparza, S., Leigh, F. A., Muzikansky, A., Park, E. R., & Plotkin, S. R. (2012). Emotional functioning of patients with neurofibromatosis tumor suppressor syndrome. *Genetics in Medicine*, *14*(12), 977–982. https://doi.org/10.1038/gim.2012.85

Theory Driven Research Project: How do autistic traits influence affirmative experiences of gender identity?

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Target journal: Archives of Sexual Behaviour

This journal has been chosen due to research being relevant to topics it covers. The journal has previously published articles pertaining to experiences of gender affirmatory treatments and the mental health of gender diverse people and the present research provides further evidence in this area.

Abstract

This research examined the experiences of gender diverse people with differing levels of autism spectrum disorder (ASD) traits in their physical and social affirmations of gender. Responses to an online survey were split into two groups and analysed according to high and low ASD trait levels. Findings revealed that no differences existed between the groups in importance and sense of passing as their identified gender. There were also no differences between groups in social belonging to gender diverse groups and gender congruence. The only significant difference found was that people with higher levels of ASD traits reported lower psychological functioning. Thus, services need to be mindful of ensuring they are autism-informed in their standard approach and recognise the additional stress that ASD can place on wellbeing. This is particularly salient when people belong to multiple minority groups where impression management occurs across an intersection of stigmatised identities.

Key words: Transgender, Autism, Affirmation, Social belonging

Introduction

Minority stress in relation to Gender Diverse and Autism Spectrum Disorder identities

Gender diverse (GD) groups are defined as people identifying as a different gender to the sex they were assigned at birth, which is experienced as gender dysphoria when their sex characteristics do not fit their identity. This contrasts with cisgender identity where people identify as the sex they were assigned at birth. Gender dysphoria has a prevalence rate of 0.55% in England and 0.40% in Wales (Office for National Statistics, 2023). Advances in protection and acceptance of GD groups have been made through the Equality Act (GOV.UK, 2010), greater visibility in the media and general public consensus of

requirements for workplace protections (Jones et al., 2018). However, GD people still experience discrimination, stigmatisation and violence both in subtle and overt forms, with subtler forms of discrimination being harder to detect and address (Swann et al., 2020). The Minority Stress Model (Meyer, 2003), has been adapted to reflect experiences of the GD population, emphasising how incidences of discrimination and violence due to gender expression and identity lead to expectations of further persecution and impact upon mental health (Hendricks & Testa, 2012). It is postulated that expectations about how others and society will behave leads people to attempt to conceal their GD identity to protect themselves from harm. However, this can contribute to internalised stigma, isolation, psychological distress, suicide, and a further stressor of non-affirmation due to balancing the enactment of their authentic identity with potential external threats (Hendricks & Testa, 2012; Rood et al., 2017). Whilst it is recognised that minority stressors can be very harmful, identification with a minority group can be protective. It enables the formation of a group identity or felt sense of belonging to the group, creates opportunities for the giving and receiving of social support and resources, and solidarity in working against prejudices and sources of threat (Branscombe et al., 1999; Crabtree et al., 2010; Hendricks & Testa, 2012). The minority stress model has also been adapted for people with Autism Spectrum Disorder (ASD) (Botha & Frost, 2020); a neurodevelopmental condition that has been identified as having a high prevalence in the GD community with individuals being 3.03-6.36 times more likely to have ASD than cisgender people (Strauss et al., 2021; Warrier et al., 2020). There are many similarities across the Minority Stress Models for GD and ASD populations (Botha & Frost, 2020; Hendricks & Testa, 2012), including stigmatisation, discrimination, poorer psychological wellbeing and concealment of minority group membership. Furthermore, should individuals be members of other minority groups, such stressors occur at multiple intersections of their identities. Therefore, GD individuals with

ASD may experience a multitude of stressors whereby there is an intersectionality of gender and neurodiverse social categorisations. This potentially creates further minority stress and requires increased effort to manage the perceptions of others to conceal or assimilate the stigmatised identities.

One way in which people manage their gender dysphoria is to seek gender affirming treatments, such as surgery or hormones (NHS, 2017). Such treatments allow individuals to take steps towards presenting as their authentic self, increase self-acceptance and facilitate greater acceptance as their identified gender by others (Hughto et al., 2020; Rood et al., 2017). However, accessing gender treatments can be difficult with long waiting lists for NHS gender clinics. In a UK study, 23% of individuals referred to a gender clinic were estimated to self-administer hormones, mostly acquired online, to achieve physical transition (Metastasio et al., 2018). Physical transition can aid broader gender affirmation by validating individuals' gender identity and treatments have been inversely related to suicidality as well as symptoms of depression, anxiety and stress (Hughto et al., 2020; Owen-Smith et al., 2018).

Social impacts of affirmation

The gender affirmation framework (Sevelius, 2013) highlights the importance of social affirmation by family, peers and society, with people who experienced rejection from their families tending to engage in riskier behaviours to feel affirmed socially. It is important to acknowledge that affirmation can occur across many domains including medical, psychological, social and legal (Smith et al., 2021). Qualitative studies have found that concealment of birth sex or GD identity are strategies used by individuals to blend in and feel accepted, with some noting that being unable to pass as their identified gender in public leads to feeling unsafe, self-conscious and suicidal (Brumbaugh-Johnson & Hull, 2019; Rood et al., 2017). Social concealment has been associated with lower self-esteem

and lack of felt authenticity in student populations (Crabtree & Pillow, 2020). It has also been highlighted that 'coming out' as GD is socially complex, with people experiencing their identity as an ongoing management process as well as a developmental process whereby strategies such as self-definition, fostering self-belief, activism and connection are instrumental in coping (Brumbaugh-Johnson & Hull, 2019; Smith et al., 2021). Those with an ASD diagnosis may experience such affirmations, concealments and disclosures as an even more stressful process due to the intersection of their GD and neurodiverse identities. Such intersection means their belonging can also be thwarted in neurotypical groups, leading to impression management of ASD identity alongside GD identity (Ai et al., 2022; Pelton et al., 2020).

Gender affirmation in the GD-ASD population

General themes of concealment, stigma and discrimination occur in Minority Stress Model adaptations for both GD (Hendricks & Testa, 2012) and ASD (Botha & Frost, 2020) populations. Qualitative literature suggests that GD individuals with ASD can struggle to fully enact their gender identity due to difficulties finding a group they feel fully affiliated with (Coleman-Smith, Smith, Milne, & Thompson, 2020; Elderton & Jones, 2011; Strang, et al., 2020). This is likely to be the result of stressors associated with ASD such as social communication, others disproportionately attributing gender dysphoria to ASD, and high expectations of life following gender confirming treatment (e.g. expecting immediate changes to appearance), which make it harder for people with ASD to integrate themselves fully into GD communities (Coleman-Smith et al., 2020; Elderton & Jones, 2011; Strang et al., 2021). Additionally, some people with ASD reported that following gender affirmative treatment, their social coping strategies did not translate well when living as their identified gender. Opportunities for socialising with GD people were reported as difficult due to noise sensitivities and locations of meet-ups (Coleman-Smith et al., 2020; Strang et al.,

2021). Such difficulties lead to further feelings of stress and difference. However, people also reported feeling a sense of ease and kinship with others due to feeling happier in their bodies and trusting that the people they socialised with were more similar to them (Strang et al., 2021). Conversely it has been highlighted that when joining groups specific to neurodiverse LGBT populations, GD individuals can still feel the groups do not cater fully for their gender experiences (Elderton & Jones, 2011). Whilst research into specific GD-ASD groups is in its infancy, such findings provide support for the minority stress model being particularly salient for people who experience an intersectionality of stigmatised identities who can find it hard to discover social groups that they fully identify with. Furthermore, it has been found that higher levels of perceived social support and lower levels of autistic traits in GD individuals have been associated with a reduction in anxiety and depression scores following gender affirmatory hormone treatments (Aldridge et al., 2021). Such findings further support accounts of GD people diagnosed with ASD experiencing difficulty affirming their identities (Coleman-Smith et al., 2020).

Summary

Existing literature has recognized that GD people who have ASD may experience greater difficulties in enacting their identities and feeling affirmed in their gender as well as experiencing an intersectionality of minority stress (Coleman-Smith et al., 2020; Elderton & Jones, 2011; Strang et al., 2021). The present study seeks to expand on this literature in order to explore whether there is a quantitative difference between the physical gender affirmation and social identification experienced between GD groups with differing levels of ASD traits. Extant literature is largely qualitative which allows for a rich insight into the experience of individuals and generation of hypotheses regarding the wider population. However, it is limited in its ability to inform clinical practice and requires quantitative testing. The aim of the present study is to provide a larger data set from which to draw

conclusions, building upon existing qualitative data in examining the experiences of people who are GD with varying degrees of ASD traits. It is also hoped this data will aid in informing 'ASD aware' gender services and clinical practice. Given the literature suggesting that GD people with ASD struggle to feel physically affirmed in their gender and can struggle to enact their identity socially, it is hypothesized that GD people with higher levels of ASD traits will experience less physical gender congruence and less affinity with a GD social identity than those with lower levels of ASD traits.

Methods

Measures

The survey included the following measures:

- ***Demographic data:*** age, gender identity, ethnicity, employment status, gender interventions, if the participant felt that passing is important to them and if they felt they did pass.

- ***ASD trait measure:*** The AQ-Short (Hoekstra et al., 2011) is a 28-item measure (α=.85) of ASD traits (Appendix B). This measure has high construct validity and criterion validity in not asking questions that may pick up on gender identity concerns. The AQ-Short has also been recognised as more suitable for research than it's lengthier counterparts, and to have high sensitivity (.94) and specificity (.91) (Hoekstra et al., 2011). A measurement of traits rather than clinical diagnosis was chosen due to it being recognised that people who are GD may not disclose ASD diagnostic status for fear of treatment denial as well as people not always receiving the diagnosis until having contact with services or not having access to appropriate services to diagnose in adulthood. Additionally this allows for different levels of traits to be examined.

- ***In-group identification:*** The Group-Level Self Definition and Self Investment Scale (Leach et al., 2008) was framed to assess GD social identification (Appendix C). The scale consists of 14 items (α=.89) scored on a seven point Likert scale (1- Strongly Disagree to 7- Strongly Agree). Higher scores indicate stronger identification. This measure uses the scales of self-definition (how similar to the group the person feels and how similar group members are perceived to be) and self-investment (how much of a bond the person feels with the group and positive feelings about group membership). Good validity has been found in the scales (Leach et al., 2008).

- ***Gender affirmation and social belonging:*** The Gender Congruence and Life Satisfaction Scale (GCLS: Appendix D) was used to assess participant's physical gender congruence through affirmations, feelings of wellbeing and life satisfaction (Jones et al., 2019). This 38 item scale has been developed and validated as an outcome measure to be used in health services to incorporate multiple GD identities. The measure was adapted to a four-point Likert scale (1- Often to 4- Never) with some items reverse scored. The adapted measure was found to be highly reliable overall (38 items, α = .92) and in relation to its two clusters: gender congruence (17 item, α = .84) and gender related mental well-being and general life satisfaction (21 items, α = .89).

Involving people with personal experience

The study was developed in collaboration with people with lived experience of both GD and ASD. The research question itself was explored and developed in consultation with a GD individual with a diagnosis of ASD and the questions and study materials were presented for group feedback in a gender service. Feedback was incorporated and actioned

as possible given questionnaire restraints, for example not comparing the results by gender and adding further trigger warning to potentially distressing sections of the questionnaire.

Participants and recruitment

An online community sample of GD participants was recruited through social media and snowball sampling (Appendix E) whereby people were encouraged to share a Qualtrics survey with others potentially interested. Inclusion criteria were identification as GD, aged over 18, resident in the UK and accessing some form of gender intervention (either through medical, non-medical, official or unofficial means). Participants were required to read an information sheet and give consent that was embedded in Qualtrics prior to accessing the survey.

Initially 280 responses were recorded; however, 127 responses were excluded due to incomplete data sets on the AQ-Short, or participants indicating they had not begun any confirmatory treatments. On the AQ-Short 83.2% (n=134) scored above the threshold of ≥65/112 suggesting they would likely meet clinical criteria for an autism diagnosis. Due to the large difference in number of participants above and below the threshold for ASD, a percentile split at the 40^{th} and 60^{th} percentiles was adopted for analysis to ensure a more even split between groups. Participants who scored between the 41^{st} and 59^{th} percentiles (AQ scores of 77-82, n= 25) were therefore excluded, giving a total participant number of 128. Participants who scored ≤77 (n=62, 48.4%) were categorised as a lower trait group, and participants scoring ≥82 (n=66, 51.6%) on the AQ-short were categorised as a higher trait group. The range of the lower trait group was 46-77 (mean= 69.35) and the higher trait group was 82-101 (mean= 88.18).

All included participants answered AQ-Short and Social Identity questions; four did not answer all questions on the GCLS, though data was included in the analysis for scales they

answered in full. The present study received ethical approval from the University of Oxford Research Ethics Committee (R79045/RE003).

Data Analytic Strategy

G* power analysis indicated that power was at an acceptable level for T-tests (β= .80) with a medium effect size (d=.5) and an acceptable level for Mann-Whitney U (β= .99) with a large effect size (d=.8). Tests of normality (Appendix F) showed that scores on measures were normally distributed, with the exception of scores on group level solidarity and centrality. Data for these measures was therefore analysed using non-parametric Mann-Whitney U.

Results

Demographics

Demographics of the two ASD trait groups can be seen in Table 1. Chi-Square analysis showed groups did not differ significantly. Participants were largely younger than 36 years old, with 71% of the low trait group and 78.8% of the higher trait group being under the age of 36, which is reflective of responses to other surveys and population statistics for GD people (GOV.UK, 2019; Nolan et al., 2019; Strader, 2022). The majority of respondents identified as male and non-binary, which was unusual given that typically a proportion of 2:1 women to men is identified in the GD community (Nolan et al., 2019).

Table 1: Demographics in percentages of the high and low trait groups

Demographic	**Category**	**Low trait levels (n=62)**	**High trait levels (n=66)**	**Chi-Square tests**
Gender	Male	40.3 (n=25)	42.4 (n=28)	
	Female	22.6 (n=14)	15.2 (n=10)	
	Non-binary	32.3 (n=20)	33.3 (n=22)	X^2 (3, N = 128) = 1.808, p = *.613*
	Other non-conforming gender identity	4.8 (n=3)	9.1 (n=6)	
Age Range	18-25	37.1 (n=23)	51.5 (n=34)	
	26-35	33.9 (n=21)	27.3 (n=18)	
	36-45	19.4 (n=12)	12.1 (n=8)	
	46-55	8.1 (n=5)	4.5 (n=3)	X^2 (5, N = 128) = 5.534, p = *.354*
	56-65	0.0 (n=0)	3.0 (n=2)	
	66-75	1.6 (n=1)	1.5 (n=1)	
Employment	Full time	51.6 (n=32)	42.4 (n=28)	
	Part time	14.5 (n=9)	15.2 (n=10)	
	Student	21.0 (n=13)	24.2 (n=16)	
	Homemaker	1.6 (n=1)	1.5 (n=1)	X^2 (5, N = 128) = 2.507, p = *.775*
	Retired	3.2 (n=2)	1.5 (n=1)	
	Unemployed	8.1 (n=5)	15.2 (n=10)	
Ethnicity	White	83.9 (n=52)	95.5 (n=63)	X^2 (3, N = 128) = 5.218, p = *.157*
	Mixed or multiple ethnic groups	8.1 (n=5)	3.0 (n=2)	
	Other ethnic group	4.8 (n=3)	1.5 (n=1)	
	Asian or Asian British	3.2 (n=2)	0.00= (n=0)	

Participants had also accessed a range of gender interventions (Tables 2 and 3) and had varying opinions on the importance and sense of passing as their identified gender (Table 4).

Table 2: Gender interventions accessed by participants

Intervention	**Low trait levels (n=62)**	**High trait levels (n=66)**	**Total (n=128)**
Prescribed medication	82.3 (n=51)	74.2 (n=49)	78.1 (n=100)
Binders or other 'non-medical' shape changers e.g. padding	75.8 (n=47)	77.3 (n=51)	76.6 (n=98)
'Top' surgery	32.3 (n=20)	22.7 (n=15)	27.3 (n=35)
Non-prescribed medication or supplements	19.4 (n=12)	21.2 (n=14)	20.3 (n=26)
Hair removal	22.6 (n=14)	15.2 (n=10)	18.8 (n=24)
Other	14.5 (n=9)	12.1 (n=8)	13.3 (n=17)
'Bottom' surgery	4.8 (n=3)	7.6 (n=5)	6.3 (n=8)
Facial cosmetic surgery	0.0 (n=0)	1.5 (n=1)	0.8 (n=1)

N.B. Participants may have had more than one intervention

Table 3: Gender interventions specified by participants under ‘other’

Intervention	**Number of participants (n)**
Hysterectomy	6
Speech/ vocal therapy	5
Therapy/ counselling	3
Name change	1
Clothes	1
Grooming products	1
Fertility preservation	1
Haircut	1
Packer	1
Laser hair removal	1

N.B. Some participants specified multiple interventions under ‘other’

Table 4: Importance of passing and felt sense of passing across groups

		Low trait levels (n=62)	High trait levels (n=66)	Total (n=128)	Chi-Square tests
Is passing important to you?	Yes	77.4 (n=48)	68.2 (n=45)	72.7 (n=93)	
	No	11.3 (n=7)	16.7 (n=11)	14.1 (n=18)	X^2 (, N = 128) = 1.391, p = .499
	Don't know	11.3 (n=7)	15.2 (n=10)	13.3 (n=17)	
Do you feel you pass?	Yes	33.9 (n=21)	31.8 (n=21)	32.8 (n=42)	
	Sometimes	37.1 (n=23)	33.3 (n=22)	35.2 (n=45)	X^2 (3, N = 128) = .824, p = .844
	No	17.7 (n=11)	24.2 (n=16)	21.1 (n=27)	
	Don't know	11.3 (n=7)	10.6 (n=7)	10.9 (n=14)	

Gender congruence

Passing

Chi-square analysis found no association between ASD traits and importance of passing χ^2 (2, N = 128)= 1.39, p=.50. Furthermore, no association was found between ASD traits and felt sense of passing χ^2 (3, N=128)= .82, p=.84. However the number of participants reporting that passing was important to them was over double the number of people who felt they did pass with 48 people saying that it was important, but only 21 saying they felt they passed as their gender.

Gender congruence and life satisfaction

An independent samples T-test was conducted to determine whether there was a difference between the gender congruence and wellbeing and life satisfaction scores of the high and

low ASD trait groups. Results indicated that there was no difference between the high (*M*=2.83, *SD*= .59) and low (*M*=2.54, *SD*= .61) ASD trait groups in overall gender congruence scores (p=.784), with a medium effect size, *d* =.60. No difference was found between overall wellbeing and life satisfaction scores of the high (*M*=2.19, *SD*= .54) and low (*M*=2.51, *SD*= .64) trait groups (p=.068), with a medium effect size, *d* =.59.

Independent T-tests examining gender congruence subscales (Table 5) determined there was a significant difference between psychological functioning of the high and low ASD trait groups (p=0.025) with people with lower traits experiencing greater psychological functioning with a medium effect size (*d=.78*). No other differences were found across subscales for gender congruence suggesting that GD people with higher levels of ASD traits do not experience less physical gender congruence than those with lower levels of ASD traits.

Table 5: Association between ASD traits and gender congruence and life satisfaction subscales

GCLS subscale	Low ASD Traits Mean (SD)	High ASD Traits Mean (SD)	*d*	p
Genitalia	2.65 (.70)	2.45 (.71)	.71	.618
Chest	2.43 (1.21)	2.20 (1.13)	1.17	.198
Other secondary sex characteristics	2.43 (.97)	2.30 (.94)	.96	.698
Social gender role recognition	2.75 (.77)	2.57 (.78)	.77	.820
Physical and emotional intimacy	2.56 (.87)	2.17 (.74)	.80	.082
Psychological functioning	2.33 (.85)	2.01 (.70)	.78	.025*
Life satisfaction	2.71 (.53)	2.45 (.51)	.52	.73

**p≤.05*

N.B. Participants who indicated that they had had 'genital surgery' on question 26 of the GCLS were excluded from the Genitalia subscale, in accordance with scoring guidelines (Jones et al., 2019).

Social belonging

Independent T-Tests found no differences between the social belonging to gender diverse groups scores of the high (*M*=39.62, *SD*= 7.65) and low (*M*=40.15, *SD*=6.83) trait groups for group level self-investment (p=.348), with a medium effect size, *d*=.726). No differences were also found between high (*M*=12.67, *SD*= 3.19) and low (*M*=12.18, *SD*= 3.44) groups in group level self-definition scores (p=.437, *d*=3.31). Results demonstrate that there is no difference between in-group identification of GD people with higher and lower levels of ASD traits.

ASD traits and domains of social identity

A Mann-Whitney U test determined there was no significant differences between the social identity subscale of solidarity for the high (*Mdn*= 14, n=66) and low (*Mdn*=14, n=62) ASD trait groups, *U*=1904.50, *z*=-.705, *p*=.481, with no effect size, r=-.062. No significant differences were also found between the social identity subscale of centrality for high (*Mdn*= 13, n=66) and low (*Mdn*=13, n=62) ASD trait groups, *U*=2038.50, *z*=-.036, *p*=.971, with no effect size, r=-0.003.

Independent samples T-Tests found no significant differences between social identification subscales of satisfaction, homogeneity or self-stereotyping, though large effect sizes were observed (Table 6). Results indicate that people with higher levels of ASD traits do not experience less affinity with a GD social identity than those with lower levels of ASD traits.

Table 6: T-tests between ASD traits and social identity subscales

Social Identity subscale	**Low ASD Traits Mean (SD)**	**High ASD Traits Mean (SD)**	***d***	**p**
Satisfaction	14.55 (3.59)	14.27 (3.97)	3.79	.331
Homogeneity	5.67 (1.86)	5.97 (1.79)	1.82	.367
Self-stereotyping	6.50 (1.96)	6.70 (1.84)	1.90	.518

Discussion

This study explored the relationship between ASD traits and physical and social gender congruence of GD people. In contrast to previous studies (GOV.UK, 2019; Strader, 2022), there was a higher response rate in people identifying as male and non-binary, which suggests a skew in the expected participants given a typically reported proportion of 2:1

women to men in the GD community (Nolan et al., 2019). However, this ratio does not incorporate identities outside of this binary and relies upon surgery clinic data. It is also worth noting that comparison to census data (Office for National Statistics, 2023) suggests the sample in the present study has higher numbers of trans male, trans female and non-binary participants compared to national data (18.52% of population trans male or trans female, 11.11% non-binary). However, people identifying as another non-conforming gender identity (4.8% in low trait group, 9.1% in high trait group) was comparable to census data (7.41%). Despite this, 44.44% of census respondents did not expand to classify their gender identity other than it being different from the sex they were assigned at birth, so conclusions regarding the representativeness of the present sample to the general population are limited. Furthermore, due to limited data on GD population demographics, it is not possible to compare representativeness of other demographic information to the GD population in the UK.

Most participants (72.7%, n=93) reported that passing as their identified gender was important to them with no difference between the importance of passing or felt sense of passing between the higher and lower ASD trait groups. However, despite this only 32.8% (n=42) felt they did pass. No differences were found between physical feelings of affirmation of GD people with higher or lower levels of ASD traits, contrary to hypothesis 1 that there would be differences. Whilst qualitative research suggests that people with ASD may have more difficulties enacting their GD identity due to the intersection of minority stress (Coleman-Smith et al., 2020; Elderton & Jones, 2011; Strang et al., 2021), these finding were not present in the current study. Additionally, contrary to hypothesis 2 that GD people with higher levels of ASD traits would experience less affinity with a GD social identity, no differences were found in social identity domains, however a significant difference was found in psychological functioning between groups. This is despite

qualitative literature highlighting the challenge of social belonging for GD people, particularly those with neurodiversity (Coleman-Smith et al., 2020; Elderton & Jones, 2011). The present findings potentially reflect an ability of people to draw on strategies used to manage one identity domain and apply them across others to reduce stressors (Smith et al., 2021) though also may suggest the extra potential burden that doing so puts on psychological wellbeing. It is however possible that as a criterion for inclusion was to be accessing some form of gender intervention, physical enactment of GD identity allowed for people to feel affirmed in their identities across the groups. Furthermore, as the participants were recruited online, it is possible that participants also utilised the internet for spaces and virtual environments whereby they could feel part of a social group without some of the stressors that offline environments present. Such benefits of online spaces for belonging and support have been recognised in both GD and ASD populations (Cipolletta et al., 2017; Jordan, 2010).

Physical and emotional intimacy and social gender role recognition also revealed no differences between the groups. Findings suggest that the experiences of GD people with varying ASD traits are not quantitatively different from each other and support the notion that group identification may be protective for individuals and lead to personal resilience in the face of other stressors (Coleman-Smith et al., 2020; Hendricks & Testa, 2012; Meyer, 2003). However, for those with higher levels of ASD traits, psychological functioning was found to be lower. It is possible that, in line with minority stress models (Botha & Frost, 2020; Hendricks & Testa, 2012) and research indicating that people with ASD work hard to manage how they are perceived in neurotypical groups (Ai et al., 2022), people with higher levels of ASD traits may exert increased effort to conceal both their gender and ASD identities. The resultant stress inherent in this might leave them at greater risk of internalised stigma and impact wellbeing (Botha & Frost, 2020; Rood et al., 2017). These

findings highlight the need to keep gender identity services ASD-informed to ensure that implications of poorer psychological wellbeing of people with ASD traits can be supported alongside consideration of gender identity needs, though also positively reflect that services do not need to be overly concerned about ASD impacting GD affirmation. The results add weight to the importance of qualitative studies to bring people's voices and nuance to the forefront of service development (Coleman-Smith et al., 2020; Elderton & Jones, 2011; Strang et al., 2021).

Study limitations

Due to 83.2% (n=134) of respondents scoring above threshold (65≤) for ASD on the AQ-Short, a median split of the data was undertaken, creating low and high trait groups for comparison. This meant that the lower trait group comprised of a mix of people who were both above and below the ASD threshold which could threaten the internal validity of the study given the large number of participants scoring above the threshold. However, this strategy allowed for people who fell just below threshold to be included and as this research considered trait levels rather than diagnosis, it was felt that this split was appropriate. Furthermore when considering ASD traits, it is reasonable to expect that people scoring more highly on the AQ-Short, are likely to experience higher levels of impairments in coping in a neurotypical environment due to having multiple domains impacted by ASD traits, in line with the initial hypotheses. Therefore, whilst conclusions are limited, given that data from 127 participants showed no significant impact of ASD traits on physical and social gender congruence, it is still reasonable to note that the initial study hypotheses were not met. Analysis of the data using ASD traits as a continuous dependent variable in a regression framework may have negated this threat to internal validity however was not conducted due to the ethics approval agreeing a group comparison study and wanting to avoid data manipulation in favour of significant results.

One limitation was not asking for confirmation of ASD diagnosis to validate the AQ-Short scores as having ASD traits is not the same as having an ASD diagnosis. However, diagnostic confirmation was deliberately omitted to negate issues of exclusion whereby people may be reluctant to disclose ASD identity and to allow for people to take part that may meet criteria for ASD but were waiting for formal ASD assessment or were unable to access this freely. Further quantitative studies with individuals with formal diagnoses of ASD are required to corroborate the findings. Furthermore, there was a large drop out in participants who were excluded due to not completing the AQ-Short or had not begun confirmatory treatments, reducing the initial participants recruited from 280 to 153 who met criteria for inclusion. Such high levels of non-completion threaten the generalisability of the study as it suggests that there may have been a sampling bias towards people who felt the topic warranted engaging with all of the questions on the survey. In order to examine whether there was any systematic differences in the demographics of people who did not complete the survey, descriptive statistics and chi-square analyses would have been appropriate to consider running.

Study implications

The present research demonstrates that there are more similarities than differences across the experiences of GD people with ASD traits and therefore services need to be mindful about making assumptions, diagnostic overshadowing, overgeneralising or excluding based on ASD diagnosis. However, as people with higher levels of traits expressed lower psychological functioning, further research needs to explore factors contributing to lower psychological functioning which might include barriers that these individuals may face in managing social inclusion, stigma and psychological distress (Ai et al., 2022; Rood et al., 2017; Strauss et al., 2021).

Future directions

Future research would benefit from exploring the intersectionality of other minority groups, such as religion, race and class, in experiences of gender and social inclusion within GD populations. This would allow further expansion on the assumption of epistemic fluency of skills being used to overcome minority stress and identify where services may further promote inclusivity. It is worth noting that comparison of the present study to census data for gender split was not possible due to the census not incorporating genders outside of the binary, despite developments in capturing transgender demographics (results not yet released to the public). Therefore it would be important for future research to also ensure that samples are representative of population proportions to improve the generalizability of findings. Future research should also explore other explanations for the poorer psychological wellbeing of GD people with high ASD traits to determine how best to address these difficulties. A further study using an ASD diagnostic split would also be advisory to see if the present results replicate when people taking part have a definitive diagnosis. Furthermore, exploration of the impact of digital spaces on social belonging for people with GD and ASD would provide a useful insight into factors that may mitigate isolation and promote group belonging.

In conclusion, the present study found no differences between GD people with high and low levels of ASD traits in their physical affirmation or social belonging, though people with higher levels of ASD traits were found to have lower psychological functioning.

References

Ai, W., Cunningham, W. A., & Lai, M.-C. (2022). Reconsidering autistic 'camouflaging' as transactional impression management. *Trends in Cognitive Sciences*, *26*(8), 631–645. https://doi.org/10.1016/j.tics.2022.05.002

Aldridge, Z., Patel, S., Guo, B., Nixon, E., Pierre Bouman, W., Witcomb, G. L., & Arcelus, J. (2021). Long-term effect of gender-affirming hormone treatment on depression and anxiety symptoms in transgender people: A prospective cohort study. *Andrology*, *9*(6), 1808–1816. https://doi.org/10.1111/andr.12884

Botha, M., & Frost, D. M. (2020). Extending the Minority Stress Model to Understand Mental Health Problems Experienced by the Autistic Population. *Society and Mental Health*, *10*(1), 20–34. https://doi.org/10.1177/2156869318804297

Branscombe, N. R., Schmitt, M. T., & Harvey, R. D. (1999). Perceiving pervasive discrimination among African Americans: Implications for group identification and well-being. *Journal of Personality and Social Psychology*, 135–149.

Brumbaugh-Johnson, S. M., & Hull, K. E. (2019). Coming Out as Transgender: Navigating the Social Implications of a Transgender Identity. *Journal of Homosexuality*, *66*(8), 1148–1177. https://doi.org/10.1080/00918369.2018.1493253

Cipolletta, S., Votadoro, R., & Faccio, E. (2017). Online support for transgender people: An analysis of forums and social networks. *Health & Social Care in the Community*, *25*(5), 1542–1551. https://doi.org/10.1111/hsc.12448

Coleman-Smith, R. S., Smith, R., Milne, E., & Thompson, A. R. (2020). 'Conflict versus Congruence': A Qualitative Study Exploring the Experience of Gender Dysphoria for Adults with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, *50*(8), 2643–2657. https://doi.org/10.1007/s10803-019-04296-3

Crabtree, J. W., Haslam, S. A., Postmes, T., & Haslam, C. (2010). Mental Health Support Groups, Stigma, and Self-Esteem: Positive and Negative Implications of Group Identification: Mental Health and Stigma. *Journal of Social Issues*, *66*(3), 553–569. https://doi.org/10.1111/j.1540-4560.2010.01662.x

Crabtree, M., & Pillow, D. (2020). Consequences of Enactment and Concealment for Felt Authenticity: Understanding the Effects of Stigma through Self-Distancing and Motive Fulfillment. *European Journal of Social Psychology*, *50*(6), 1227–1247.

Elderton, A., & Jones, C. (2011). Finding a safe place to explore sexual identity. *Learning Disability Practice*, *14*(5), 14–18. https://doi.org/10.7748/ldp2011.06.14.5.14.c8559

GOV.UK. (2010). *Equality Act 2010* [Text]. Statute Law Database. https://www.legislation.gov.uk/ukpga/2010/15/contents

GOV.UK. (2019, February 7). *National LGBT Survey: Summary report*. National LGBT Survey: Summary Report. https://www.gov.uk/government/publications/national-lgbt-survey-summary-report/national-lgbt-survey-summary-report

Hendricks, M. L., & Testa, R. J. (2012). A conceptual framework for clinical work with transgender and gender nonconforming clients: An adaptation of the Minority Stress Model. *Professional Psychology: Research and Practice*, *43*(5), 460–467. https://doi.org/10.1037/a0029597

Hoekstra, R. A., Vinkhuyzen, A. A. E., Wheelwright, S., Bartels, M., Boomsma, D. I., Baron-Cohen, S., Posthuma, D., & van der Sluis, S. (2011). The Construction and Validation of an Abridged Version of the Autism-Spectrum Quotient (AQ-Short). *Journal of Autism and Developmental Disorders*, *41*(5), 589–596. https://doi.org/10.1007/s10803-010-1073-0

Hughto, J. M. W., Gunn, H. A., Rood, B. A., & Pantalone, D. W. (2020). Social and Medical Gender Affirmation Experiences Are Inversely Associated with Mental Health Problems in a U.S. Non-Probability Sample of Transgender Adults. *Archives of Sexual Behavior*, *49*(7), 2635–2647. https://doi.org/10.1007/s10508-020-01655-5

Jones, B. A., Bouman, W. P., Haycraft, E., & Arcelus, J. (2019). The Gender Congruence and Life Satisfaction Scale (GCLS): Development and validation of a scale to measure outcomes from transgender health services. *International Journal of Transgenderism*, *20*(1), 63–80. https://doi.org/10.1080/15532739.2018.1453425

Jones, P. E., Brewer, P. R., Young, D. G., Lambe, J. L., & Hoffman, L. H. (2018). Explaining Public Opinion toward Transgender People, Rights, and Candidates. *Public Opinion Quarterly*, *82*(2), 252–278. https://doi.org/10.1093/poq/nfy009

Jordan, C. J. (2010). Evolution of Autism Support and Understanding Via the World Wide Web. *Intellectual and Developmental Disabilities*, *48*(3), 220–227. https://doi.org/10.1352/1934-9556-48.3.220

Leach, C. W., van Zomeren, M., Zebel, S., Vliek, M. L. W., Pennekamp, S. F., Doosje, B., Ouwerkerk, J. W., & Spears, R. (2008). Group-level self-definition and self-investment: A hierarchical (multicomponent) model of in-group identification. *Journal of Personality and Social Psychology*, *95*(1), 144–165. https://doi.org/10.1037/0022-3514.95.1.144

Metastasio, A., Negri, A., Martinotti, G., & Corazza, O. (2018). Transitioning Bodies. The Case of Self-Prescribing Sexual Hormones in Gender Affirmation in Individuals Attending Psychiatric Services. *Brain Sciences*, *8*(5), Article 5. https://doi.org/10.3390/brainsci8050088

Meyer, I. H. (2003). Prejudice, Social Stress, and Mental Health in Lesbian, Gay, and Bisexual Populations: Conceptual Issues and Research Evidence. *Psychological Bulletin, 129*(5), 674–697. https://doi.org/10.1037/0033-2909.129.5.674

NHS. (2017, October 23). *Gender dysphoria*. Nhs.Uk. https://www.nhs.uk/conditions/gender-dysphoria/

Nolan, I. T., Kuhner, C. J., & Dy, G. W. (2019). Demographic and temporal trends in transgender identities and gender confirming surgery. *Translational Andrology and Urology, 8*(3), Article 3. https://doi.org/10.21037/tau.2019.04.09

Office for National Statistics. (2023, January 6). *Gender identity, England and Wales: Census 2021*. Census 2021: Gender Identity, England and Wales. https://www.ons.gov.uk/peoplepopulationandcommunity/culturalidentity/genderidentity/bulletins/genderidentityenglandandwales/census2021#gender-identity

Owen-Smith, A. A., Gerth, J., Sineath, R. C., Barzilay, J., Becerra-Culqui, T. A., Getahun, D., Giammattei, S., Hunkeler, E., Lash, T. L., Millman, A., Nash, R., Quinn, V. P., Robinson, B., Roblin, D., Sanchez, T., Silverberg, M. J., Tangpricha, V., Valentine, C., Winter, S., … Goodman, M. (2018). Association Between Gender Confirmation Treatments and Perceived Gender Congruence, Body Image Satisfaction, and Mental Health in a Cohort of Transgender Individuals. *The Journal of Sexual Medicine, 15*(4), 591–600. https://doi.org/10.1016/j.jsxm.2018.01.017

Pelton, M. K., Crawford, H., Robertson, A. E., Rodgers, J., Baron-Cohen, S., & Cassidy, S. (2020). Understanding Suicide Risk in Autistic Adults: Comparing the Interpersonal Theory of Suicide in Autistic and Non-autistic Samples. *Journal of Autism and Developmental Disorders, 50*(10), 3620–3637. https://doi.org/10.1007/s10803-020-04393-8

Rood, B., Maroney, M., Puckett, J., Berman, A., Reisner, S., & Pantalone, D. (2017). Identity concealment in transgender adults: A qualitative assessment of minority stress and gender affirmation. *American Journal of Orthopsychiatry*, *87*, 704–713. https://doi.org/10.1037/ort0000303

Sevelius, J. M. (2013). Gender Affirmation: A Framework for Conceptualizing Risk Behavior Among Transgender Women of Color. *Sex Roles*, *68*(11), 675–689. https://doi.org/10.1007/s11199-012-0216-5

Smith, G., Robertson, N., & Cotton, S. (2021). Transgender and gender non-conforming people's adaptive coping responses to minority stress: A framework synthesis. *Nordic Psychology*, *0*(0), 1–21. https://doi.org/10.1080/19012276.2021.1989708

Strader, E. (2022). Demographics of Transgender People and Transition-Related Healthcare Policies Across the European Union. In A. K. Baumle & S. Nordmarken (Eds.), *Demography of Transgender, Nonbinary and Gender Minority Populations* (pp. 211–237). Springer International Publishing. https://doi.org/10.1007/978-3-031-06329-9_8

Strang, J. F., Knauss, M., van der Miesen, A., McGuire, J. K., Kenworthy, L., Caplan, R., Freeman, A., Sadikova, E., Zaks, Z., Pervez, N., Balleur, A., Rowlands, D. W., Sibarium, E., Willing, L., McCool, M. A., Ehrbar, R. D., Wyss, S. E., Wimms, H., Tobing, J., … Anthony, L. G. (2021). A Clinical Program for Transgender and Gender-Diverse Neurodiverse/Autistic Adolescents Developed through Community-Based Participatory Design. *Journal of Clinical Child & Adolescent Psychology*, *50*(6), 730–745. https://doi.org/10.1080/15374416.2020.1731817

Strauss, P., Cook, A., Watson, V., Winter, S., Whitehouse, A., Albrecht, N., Wright Toussaint, D., & Lin, A. (2021). Mental health difficulties among trans and gender diverse young people with an autism spectrum disorder (ASD): Findings from

Trans Pathways. *Journal of Psychiatric Research*, *137*, 360–367. https://doi.org/10.1016/j.jpsychires.2021.03.005

Swann, G., Stephens, J., Newcomb, M. E., & Whitton, S. W. (2020). Effects of Sexual/Gender Minority-and Race-Based Enacted Stigma on Mental Health and Substance Use in Female Assigned at Birth Sexual Minority Youth. *Cultural Diversity & Ethnic Minority Psychology*, *26*(2), 239–249. https://doi.org/10.1037/cdp0000292

Warrier, V., Greenberg, D. M., Weir, E., Buckingham, C., Smith, P., Lai, M.-C., Allison, C., & Baron-Cohen, S. (2020). Elevated rates of autism, other neurodevelopmental and psychiatric diagnoses, and autistic traits in transgender and gender-diverse individuals. *Nature Communications*, *11*(1), Article 1. https://doi.org/10.1038/s41467-020-17794-1

Executive Summary of Theory Driven Research Project

The research looked at how different levels of autistic traits influence how people experience their gender identity and social belonging. Existing literature suggested that gender diverse (GD) people with a diagnosis of autism spectrum disorder (ASD) may struggle more to enact and feel affirmed in their gender than those without ASD (Coleman-Smith et al., 2020; Elderton & Jones, 2011; Strang et al., 2021). The current research sought to further investigate if there were quantitative differences between the affirmation and social identifications experienced by people who were GD with different levels of ASD traits. Current literature is mainly qualitative and so provides a rich narrative and insight into the experiences of GD people, though due to this is limited in its generalisation to the wider GD population and requires quantitative testing. The current study aimed to get a larger data set to draw conclusions and build upon existing data. It is also hoped that the present data will aid in informing gender services and clinical practice to be more 'ASD aware'.

The study examined the following hypotheses:

1. GD people with higher levels of ASD traits will experience less physical gender affirmation than those with lower levels of ASD traits.
2. GD people with higher levels of ASD traits will experience less affinity with a GD social identity than those with lower levels of ASD traits.

Methods

An online survey was used to capture the responses of a sample of GD participants who were over the age of 18, lived in the UK and were accessing a gender intervention of some form. The survey and research was developed with the input of people with lived experience of GD and ASD. The survey included demographic data, a measure of ASD

traits (Hoekstra et al., 2011), a group identification scale to assess social belonging (Leach et al., 2008) and a gender congruence scale to assess physical affirmation and life satisfaction (Jones et al., 2019).

Results

Initially 280 responses were recorded; however, 125 responses were excluded due to incomplete data sets, or participants indicating they had not begun any gender treatments. On the ASD scale, 83.2% (n=134) scored above the threshold of ≥65/112 suggesting they would likely meet clinical criteria for an autism diagnosis. Due to the large difference in number of participants above and below the threshold for ASD, analysis used a percentile split at the 40^{th} and 60^{th} percentiles to get a more even split between groups, giving a total participant number of 128. Participants were categorised as being in the low trait group or the higher trait group.

Analysis showed no differences between the groups in importance and sense of passing as their identified gender. There were also no differences between groups in social belonging and gender affirmation. The only differences found were that people with higher levels of ASD traits reported lower psychological functioning. Of the people surveyed, 73% said that passing as their identified gender was important to them, but only 33% thought that they did pass as their gender. People also reported having a range of gender interventions, with 20% of people saying that they had taken non-prescribed medications or supplements to aid their gender affirmation.

Conclusions

The data showed that there were more similarities between GD people with differing levels of ASD traits than there were differences, though people with higher levels of ASD traits struggle more in their psychological functioning than those with lower levels of traits. It is

therefore important that healthcare professionals are aware that GD people with ASD may need higher levels of support to achieve life satisfaction. It was also crucial to note than people place a lot of importance on people perceiving them as their identified gender and 20% reported taking substances that were not medically prescribed to achieve this and so interventions that are timely are key to reduce people's sense of dysphoria and increase their safety.

Connecting Narrative

904 words

Initially, I was not sure how related my projects really were, though as my knowledge has increased over the course, I have come to realise that I have grown as a clinician and researcher with each one. I have become more confident in my application and critical evaluation of research as well as developing a secure foundation of understanding from which to try new skills and methods. As my clinical skills have grown, I have also realised that whilst on the surface, the projects cover three different areas of psychology, they all have a common theme of health, wellbeing and responsivity to patient needs. At the start of this process, I would have still seen the projects as discrete and separate, however with the development of my clinical skills alongside the research, I can see how application of knowledge and research to one area is transferrable to the skills required in another, for example, stakeholder involvement, identification of basic areas that remain problematic, and examining the strength of the evidence for the population.

Working on my three projects for the last three years has allowed me to develop my research skills and find a greater appreciation for the process and application of research. The service improvement project, focussing on hearing interventions in people with neurofibromatosis type two, pushed me to utilise content analysis, a qualitative method that I had not previously done. The process of working with stakeholders to develop the research enabled me to appreciate how invested people who benefit from the research can be and encouraged me to ensure that the project was comprehensive and contained recommendations that were logical and achievable for the service to implement.

Working on the theoretically driven research project, examining the experiences of physical and social congruence of gender diverse people allowed me to build on the skills

that the service improvement project had produced. I felt more able to engage with stakeholders to ensure that their considerations were given to the project as well as being more confident to say no when suggestions would not be realistic or went against what other parties had contributed. In recruiting to this project I was struck by how strongly invested gender diverse people were and as a result I was able to recruit quickly to the study. This study also taught me to be flexible in how I approached data sets that were skewed and ways to problem solve that were statistically viable, without adding bias or invalidating the data. It is fair to say that I have never been a 'maths person', though working on the theory driven project allowed me to grow in confidence that I understand it more than I expected to. I was particularly struck by this when meeting with a statistician and they agreed with my approach when I told them my plan for analysis, something that I was not expecting to occur due to my own perceptions of my abilities. Working on the theory driven research project also showed me that how in the face of seemingly cold data; I was able to keep my empathy for the difficulties that my participants faced. I noticed that the realisation that 20% of my sample was using non-prescribed medication or supplements to address their physical appearance brought up sadness and concern in me for the experiences and dangers that my participants risk in their lives. I also noticed that when generally my results found no differences between high and low trait groups, I was heartened that the research may lead to a greater understanding that people are more alike than they are different, something that feels often forgotten in how people are pathologised.

My final project of the three was my literature review pertaining to psychoeducational approaches to non-epileptic seizures. The process of this review felt sluggish and I am still trying to understand if this was due to it being the final project of the three, or if this was to do with the process itself. It is fair to say that this project took more developing than the

others, with multiple searches being required to ensure that sufficient numbers papers would be found that met inclusion criteria. This process aided my ability to search the literature and consulting with a librarian on this was invaluable to ensure that I fully understood how to search. In discussion with my supervisor, it was decided that a scoping review would be most appropriate and I have been excited to have the opportunity to do a review that allowed for more flexibility in how papers were synthesised and reported on, despite the process of searching for the papers to be the same as other reviews done by my colleagues. This project also taught me the value of working with other researchers to collaborate and provide assistance to each other in assessing papers for inclusion of each other's respective studies. Additionally, the sharing of resources and knowledge process between trainee's created a sense of shared camaraderie that was instrumental in finding the energy to keep going when tasks seemed never ending.

In conclusion, my research and clinical experiences throughout the course have given me a new and greater appreciation for the epistemic fluency of skills in psychology and their application. I have been fortunate to have been given many opportunities for development and learning, and feel that the support of fellow trainee's has been invaluable throughout the process.

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I would like to thank my fellow trainees, friends, family and partner. Moral support goes a long way when conducting research and such camaraderie is invaluable, as is someone to cook dinner when you are in the middle of a write up!

Finally, I would like to thank my rabbits who have provided light entertainment, procrastination and often decided to make an appearance in research meetings. I am also hoping that by thanking them now, they may keep their teeth away from this thesis when it is bound (I live in hope).

Research Appendices

Scoping review Appendices

Appendix A: Target journal (Seizure- European Journal of Epilepsy) instructions for authors, taken from https://www.elsevier.com/journals/seizure-european-journal-of-epilepsy/1059-1311/guide-for-authors:

Seizure - European Journal of Epilepsy publishes the following types of article:

1.1 Peer-reviewed articles

a. Full reviews.

Seizure welcomes comprehensive reviews on all subjects relating to epilepsy and other seizure disorders. Authors planning/proposing are invited to discuss their ideas with Editor-in-Chief prior to submission. Full reviews should be preceded by an abstract. Full reviews should not exceed 7,000 words, include no more than 6 figures or tables and 150 references.

Submission checklist

You can use this list to carry out a final check of your submission before you send it to the journal for review. Please check the relevant section in this Guide for Authors for more details.

Ensure that the following items are present:

One author has been designated as the corresponding author with contact details:

• E-mail address

• Full postal address

All necessary files have been uploaded:

Manuscript:

• Include keywords

• All figures (include relevant captions)

• All tables (including titles, description, footnotes)

• Ensure all figure and table citations in the text match the files provided

• Indicate clearly if color should be used for any figures in print

Graphical Abstracts / Highlights files (where applicable)

Supplemental files (where applicable)

Further considerations

• Manuscript has been 'spell checked' and 'grammar checked'

• All references mentioned in the Reference List are cited in the text, and vice versa

• Permission has been obtained for use of copyrighted material from other sources (including the Internet)

• A competing interests statement is provided, even if the authors have no competing interests to declare

• Journal policies detailed in this guide have been reviewed

• Referee suggestions and contact details provided, based on journal requirements

• A completed reporting quality checklist from The EQUATOR Network (https://www.equator-network.org/),appropriate for the type of submission or a declaration why such a checklist does is not suitable for the manuscript being submitted (see section "Reporting Guidelines and Checklists" below for details).

Appendix B: Names for non-epileptic seizures:

- Non-epileptic seizures
- Non-epileptic attack disorder
- Non-epileptic paroxysmal event
- Non-epileptic event

- Non-epileptic attack
- Pseudoseizure
- Psychogenic seizure
- Functional seizure
- Dissociative seizure

Appendix C: Previous systematic reviews into psychoeducation for NES

Review	Content
The efficacy of psychoeducation interventions for psychogenic non-epileptic seizures: a systematic review Registered on PROSPERO as completed not published February 2019 (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=112122)	Focusses only on seizure reduction, not on mental health, quality of life or psychoeducation content. Attempts have been made to contact the researchers about this study with limited success. Currently it is fair to conclude that it is not planned to be published and it is not available on the university archives.
A systematic review identifying what factors are associated with patient reported health related quality of life in individuals experiencing psychogenic non-epileptic seizures Registered on PROSPERO February 2021, incomplete. (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=234722)	Focusses on influencing factors of quality of life in NES, not psychoeducational impact on quality of life.
A systematic review of studies comparing the psychological characteristics of functional neurological disorders Registered in PROSPERO February 2019, incomplete. (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=123555)	Focusses on any functional neurological disorder, not just NES. Examines psychological characteristics of individuals not an intervention.
Psychological treatments for non-epileptic seizures in children and adolescents. Registered on PROSPERO December 2020, incomplete. (https://www.crd.york.ac.uk/prospero/display_record.	Compares a variety of treatments, including psychoeducation, for children and adolescents only. Main focus is seizure frequency and secondary outcomes are quality of life. No mention of

php?RecordID=208900)	mental health outcomes.
Quality of life in patients with epilepsy and psychogenic nonepileptic seizures: a systematic review Registered on PROSPERO July 2020, incomplete. (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=188620)	Comparison of quality of life in people with NES and epilepsy and reasons for this difference, not on impacts of treatment.
Update on psychological interventions for adults with dissociative seizures: a systematic review Registered on PROSPERO October 2020, incomplete. (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=207281)	Focusses only on adults and on any psychological intervention, not specific to psychoeducation. Search strategy includes difference databases to the current study.

Appendix D: Search Strategy

The search strategy was developed with a librarian (Appendix C). The search strategy for MEDLINE was as follows:

1. ("functional neurological disorder*" or "FND" or "functional neurological symptom disorder*" or "functional movement disorder*" or "functional disorder*" or "nonepileptic seizure*" or "non epileptic seizure*" or "nonepileptic attack disorder*" or "non epileptic attack disorder*" or "non epileptic paroxysmal event*" or "nonepileptic paroxysmal event*" or "non epileptic event*" or "pseudoseizure*" or "pseudo seizure*" or "psychogenic seizure*" or "psychogenic nonepileptic seizure*" or "psychogenic non epileptic seizure*" or "functional seizure*" or "PNES" or "dissociative seizure*" or "conversion disorder*or motor disorder*" or NEAD).ti,ab.
2. ("psychoed*" or "psycho ed*" or "education*").ti,ab.
3. 1 and 2

A similar search was adapted for other databases.

SIP Appendices

Appendix A: Target journal (Psychology, Health and Medicine) instructions for authors, taken from https://www.tandfonline.com/action/authorSubmission?show=instructions&journalCode=cphm20:

Preparing Your Paper

Structure

Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

Word Limits

Please include a word count for your paper.

A typical paper for this journal should be no more than 3,000 words.

Papers for our online-only Methodological issue should be no more than 4,000 words. This limit does not include tables, references, figure captions and endnotes.

Style Guidelines

Please refer to these quick style guidelines when preparing your paper, rather than any published articles or a sample copy.

Any spelling style is acceptable so long as it is consistent within the manuscript.

Please use single quotation marks, except where ‘a quotation is “within” a quotation’. Please note that long quotations should be indented without quotation marks.

Formatting and Templates

Papers may be submitted in Word or LaTeX formats. Figures should be saved separately from the text. To assist you in preparing your paper, we provide formatting template(s).

Word templates are available for this journal. Please save the template to your hard drive, ready for use.

A LaTeX template is available for this journal. Please save the LaTeX template to your hard drive and open it, ready for use, by clicking on the icon in Windows Explorer.

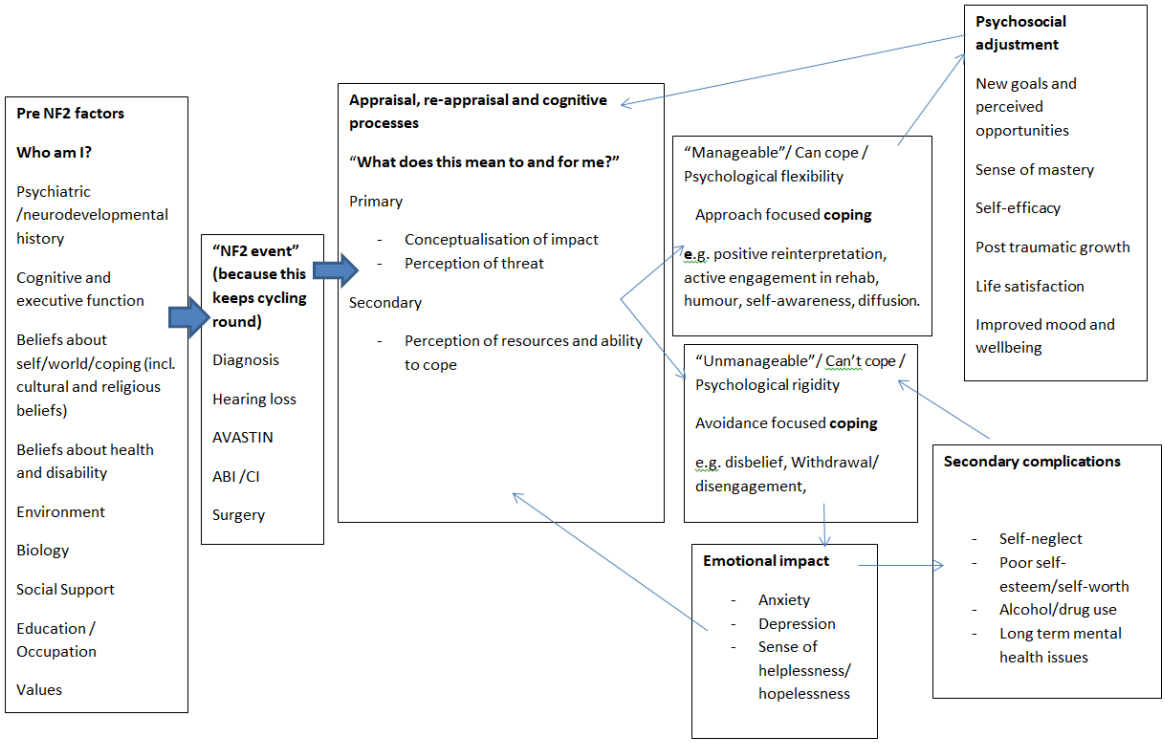
If you are not able to use the template via the links (or if you have any other template queries) please contact us here.

References

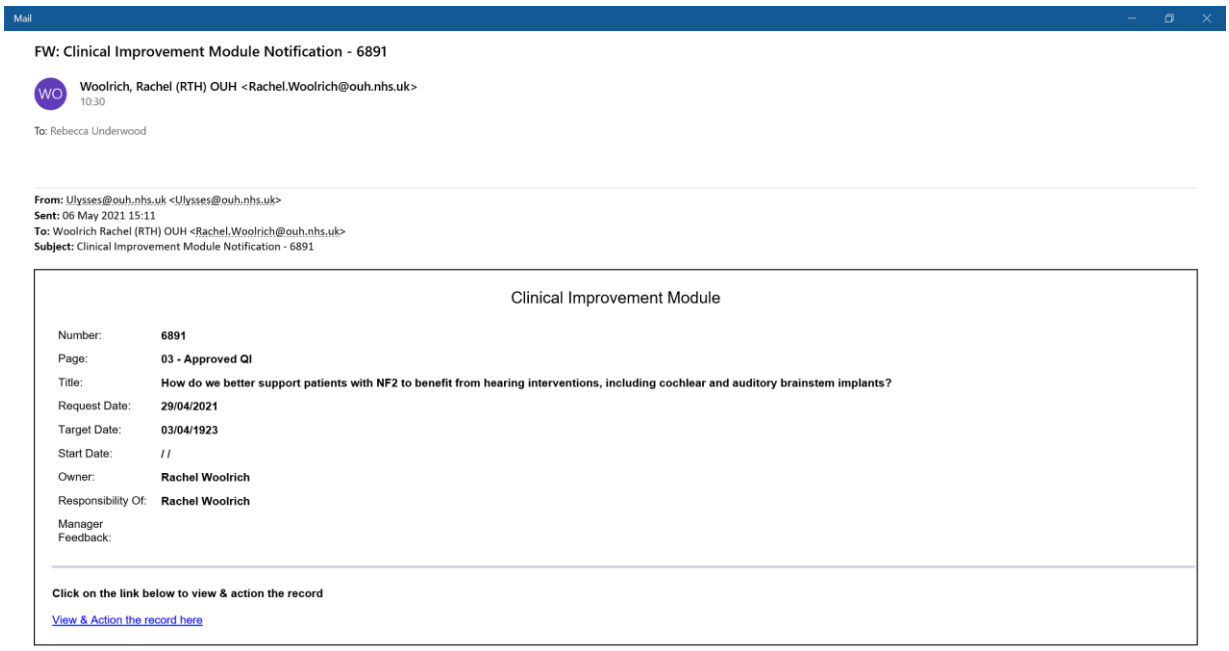
Please use this reference guide when preparing your paper.

An EndNote output style is also available to assist you.

Appendix B: A proposed model for understanding psychosocial adjustment to NF2 (adapted from Kennedy & Duff, 2005, with permission)



Appendix C: NHS Audit approval



Mail

FW: Clinical Improvement Module Notification - 6891

WO Woolrich, Rachel (RTH) OUH <Rachel.Woolrich@ouh.nhs.uk>
10:30

To: Rebecca Underwood

From: Ulysses@ouh.nhs.uk <Ulysses@ouh.nhs.uk>
Sent: 06 May 2021 15:11
To: Woolrich Rachel (RTH) OUH <Rachel.Woolrich@ouh.nhs.uk>
Subject: Clinical Improvement Module Notification - 6891

Clinical Improvement Module

Number:	**6891**
Page:	**03 - Approved QI**
Title:	**How do we better support patients with NF2 to benefit from hearing interventions, including cochlear and auditory brainstem implants?**
Request Date:	**29/04/2021**
Target Date:	**03/04/1923**
Start Date:	/ /
Owner:	**Rachel Woolrich**
Responsibility Of:	**Rachel Woolrich**
Manager Feedback:	

Click on the link below to view & action the record

View & Action the record here

Appendix D: Survey questions

Section 1: About You

1. Do you have a diagnosis of Neurofibromatosis Type 2 (NF2)?
Please tick the appropriate box

- Yes
- No
- Other – e.g. schwannomatosis
- I don't know

2. At what age were you diagnosed with NF2?

If you are unsure, please give your best estimate in years

2a. What year was your diagnosis?

3. How old are you?

Please tick the appropriate box

- 18-25 years
- 26-35 years
- 36-45 years
- 46-55 years
- 56-65 years
- 66-75 years
- 76 years and over

4. How would you describe your gender?

Please tick the appropriate box

- Male
- Female
- Non-binary

- Other non-conforming gender identity

5. Do you have a family member who also has an NF2 diagnosis?

Please tick the appropriate box

- Yes
- No
- Don’t know

6. In addition to the Oxford team, have you ever received support from any other NF2 team (e.g. London, Cambridge or Manchester)?

Please tick the appropriate box

- Yes
- No

7. How has NF2 impacted on you? Please tick all that apply

- Hearing loss/becoming deafened
- Reduced vision/loss of sight
- Other sensory loss
- Facial palsy
- Impaired balance
- Restricted mobility
- Mental health/self-esteem
- Pain
- Tiredness/ fatigue
- Other (please expand in next question)

8. Please tell us a bit more how NF2 has impacted on you.

9. Does anyone else in your family have hearing difficulties?
Please tick the appropriate box

- Yes
- No
- Don't know

9a. What Impact did your family member's hearing difficulties have on your own experience of hearing loss?

Section 2: Hearing

This section focusses on what you have been offered for your hearing:

10. Have you been offered any of the following to help with your hearing? (please rate all that apply, including historical interventions)

	Not been offered	Offered to me but I declined	Offered to me and I took it up
Hearing aid			
Cochlear Implant			
Auditory brainstem implant			

11. Of these, which are you currently using?

	Currently using	Stopped Using	Not applicable as I have never used this
Hearing aid			
Cochlear Implant			
Auditory brainstem implant			

12. Can you explain what influenced your decision to decline or take up these treatments for hearing loss?

13. If you have stopped using any of these, can you tell us a bit more about what influenced this decision to stop?

14. What other support/information have you been given **by the NF2 team** to support your hearing/communication?

	Yes	No	I have been given this but not by the NF2 team
Advice or information about charity support e.g. Can you Hear Us, Nerve Tumours UK			
Advice or information about lip reading classes			
Advice or information about sign language course			
Offered interpreter for clinic appointments			
Given information regarding voice text/audiotyping apps for phones			
Advice or information about Sensory support services/Sensory impairment team			
Other (please state below)			

Other:

14a. If you have got information from elsewhere, could you please share where you got it from?

15. Have you received any support from other sources? Please tick all that apply

- Other NHS teams
- NF2 community online
- Family/friends
- Sought information myself
- Charity/3rd Sector If yes which? (please state below)
- Sensory support service /sensory impairment team
- I have not received any support from other sources

Section 3: The rest of this questionnaire focuses on the experience of hearing loss

16. Please tick the appropriate boxes

	Strongly Agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Don't know
My experience of hearing loss is in line with what I expected to happen.						
I am satisfied with the amount of information I was given by the NF2 team about hearing loss						
The information the NF2 team have given me about hearing loss was easy to understand						
The information the Nf2 team have given me about hearing loss was relevant and helpful to me.						

I was given enough emotional support to adjust to the impact of hearing loss						

Hearing aids:

17a. Have you used a hearing aid with the NF2 Team? If No, please skip to section about Cochlear Implants and Auditory Brain Stem Implants (question 18)

- Yes
- No

17.b Your experience of deciding to have a hearing aid:

	Strongly Agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Don't know
I had time to think about the information I was given before making a decision						
I made the decision based on scientific and medical information that I was given						
I made the decision based on experiences of other people I know						
I made the decision based on logistical factors such as family and ability to travel						

to appointments						
I made the decision on gut instincts						
I was concerned about people being able to see that I was wearing a hearing device						
I was given information about the possible risks and benefits to aid my decision						

17c. Experience of the process of having a hearing aid

	Strongly Agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Don't know
I was given enough information about the procedure of having a hearing aid fitted.						
I felt supported to make the decision to have this						
I understood the importance of wearing my device and						

practicing listening with my device						
I felt ready and able to take part in the rehabilitation process (attend appointments, device adjustment, wearing the device in different settings)						
My hearing aid has helped me access sound and helped my communication						
Overall I am satisfied with the outcome of having a hearing aid						

17d. Did the hearing aid meet your expectations? Why/ why not?

17e. Do you have any comments about outcome or process of your hearing aid intervention?

17f. Would you recommend having a hearing aid to someone else with NF2?

Please tick the appropriate box

- o Yes
- o No
- o Unsure

17g. Please give reasons for your answer

18a. Have you used a **cochlear implant or auditory brain stem implant** with the NF2 Team? Please tick the appropriate box

If you answered no, please skip to the section about how the NF2 team can improve in the future (on the last page).

- o Cochlear Implant
- o Auditory Brain Stem Implant
- o No

18b. Your experience of deciding to have a cochlear implant or auditory brain stem implant:

	Strongly Agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Don't know
I had time to think about the information I was given before making a decision						
I made the decision based on scientific and medical information that I was given						
I made the decision based on experiences of other people I know						
I made the decision based						

on logistical factors such as family and ability to travel to appointments						
I made the decision on gut instincts						
I was concerned about people being able to see that I was wearing a hearing device						
I was given information about the possible risks and benefits to aid my decision						

18c. Experience of the process of having a cochlear implant or auditory brain stem implant:

	Strongly Agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Don't know
I was given enough information about the surgical procedure.						
I felt supported by the NF2 team/auditory implant team to make the decision						

to have this.						
I understood the importance of wearing my device and practicing listening with my device						
I felt ready and able to take part in the rehabilitation process (attend appointments, device adjustment, wearing the device in different settings)						
This device has helped me access sound and helped my communication generally						
Overall I am satisfied with the outcome of having this device						

18d. Did the Cochlear Implant or Auditory Brain Stem Implant meet your expectations? Why/ why not?

18e. Do you have any comments about outcome or process of your Cochlear Implant/ Auditory Brain Stem Implant?

18f. What impact has the Cochlear Implant/ Auditory Brain Stem Implant had on your understanding of speech and communication?

Please tick the appropriate box

- o Made it much harder
- o Made it somewhat harder
- o No difference

- Made it somewhat easier
- Made it much easier

18g. Would you recommend having a CI/ABI to someone else with NF2?

Please tick the appropriate box

- Yes
- No
- Unsure

18h. Please give reasons for your answer

This section focusses on what the NF2 team can do to improve in future

19. Overall, is there anything else the NF2 team could do to support you with your hearing loss and accessing hearing interventions?

20. Any other comments?

Appendix E: Stages of content analysis (Kyngäs, 2020)

Stage	**Process**
Data familiarisation	Reading through the data multiple times
Selection of unit of analysis	Broad themes identified as unit of analysis
Analysis of open codes to form sub-themes	Open codes identified by reading the data sentence by sentence (e.g. 'Worries')
Comparison of similarities and differences between open codes to inform grouping	Grouping of codes into broader themes
Checking similarities and groupings identified reflect the data set	Discussion between researchers to compare initial codings and groupings
Grouping together of main concepts	Discussion yielded an agreement of grouping concepts under the research questions they linked to (psychological wellbeing and hearing interventions)
Reporting of results	Use of text and tables to contextualise codes and themes

Appendix F: Development of codes

Appendix F1: Development of themes for question: How do patients with NF2 experience hearing loss and the support they receive from the NF2 team?

		Subtheme	**Theme**
Raw data →	*Open codes→*	*All encompassing* *Reminders of condition* *Burden of treatment* *Physical impact* *Interference with everyday activities*	*Burden of physical symptoms: "What a cruel trick our bodies play on us"*
		Overshadows experiences *Unpredictable nature of the condition* *Impact on confidence* *Shame around hearing loss* *Impact on daily activities* *Emotional impact of the unknown* *Worries about the impact on others* *Witnessing family in distress* *Worry about family/ future* *Sense of loss*	*Emotional burden: "Thinking about it makes me feel bad"*
		Feeling isolated *Impact on social life* *Impact on ability to connect with others*	*Social impact and withdrawal: "I often feel left out"*
		Multiple things to adapt to *Increased support from others* *Problem solving and supporting each other together* *Increased empathy* *Able to prepare for future* *Coping better with time*	*Impact on flexibility and adaptation to cope : "I've been able to adapt, but certain scenarios are still difficult"*
		Impact on confidence *Shame around hearing loss*	*Impact on identity and confidence: "It feels like sometimes there are no parts of me which are not affected"*
		Impact on work *Worry about discrimination at work*	*"Career progression has been significantly impacted"*

		Support at work	
		Not impacted much	*Sense of being lucky with NF2 as a whole: "I am lucky in that my NF2 is both mild and mosaic"*
		Other things aside from hearing loss impacting	*Hearing loss is not the biggest issue: "After surgical removal of schwannomas the hearing loss was not the biggest of my problems"*

Appendix F2: Development of themes for question: How do patients with NF2 experience hearing interventions in terms of information provision, decision making and general support from the team?

		Subtheme	**Theme**
Raw data →	*Open codes→*	*Got positive effects* *Used in aid of existing strategies* *Use of other methods to manage* *Initial expectations not met but happy with an adjusted view* *Exceeded expectations* *Helped with hearing*	*Hearing aid: Meeting or exceeding expectations: "Hearing aids are the closest thing to a miracle that I can imagine" / Successes of CI/ABI*
		Changing device for hearing need *Intervention taken with hope of better sound quality* *Intervention taken to improve mood* *Intervention taken to reduce burden on daily tasks* *Intervention to aid social interactions*	*Hearing aid as vital for functioning: "I would not have been able to function if I did not take it up."*
		Initial lack of use due to sound not being as hoped *Expectations not met as strongly as hoped* *Did not meet expectations* *Disappointment expressed it is not the same as hearing*	*Hearing aid: Not meeting expectations: "the sound quality was poor and music, especially, was so distorted I couldn't bear it"/ Limitations of CI/ABI*

Feel other methods are more helpful	
Visible difference *Desire to appear normal* *Sense of being disabled*	*Fear of drawing attention to difference: "I wanted to appear more normal"*
Finding the device physically uncomfortable *Technical issues* *Sense it can be difficult to use* *Worry about negative impact on hearing of intervention*	*Practical concerns: "Find it too bulky"*
Helpful for work and social life	*Social benefits of the hearing aid: "I feel a little less cut off from the world when I use them"*
Took up intervention on advice that it may help	*Trust in professional opinion: "I have never declined to try or consider positioned options"*
Intervention stopped working	*Deterioration in hearing leading to ceasing use: "it didn't work anymore"*
Worry about negative impact on hearing of intervention *Reluctance to try intervention due to unknown outcome* *Reluctance for surgery* *Sense that CI/ ABI is a last resort* *Not able to accept the hearing aids until larger interference on life* *Needing to get to a point of being worse for CI to be helpful*	*Perceptions of physical risk of CI/ABI*

Appendix F3: Development of themes for question: What improvements would patients with NF2 like to see in terms of support with hearing loss and interventions?

		Subtheme	**Theme**
Raw data →	*Open codes→*	*Input offered from NF2 team* *Praise for the NF2 team*	*Complement for the team*
		Unsure when stuff is mentioned, how to access it *More explicit signposting* *Need for clearer information about what NF2 is* *Desire for tips for hearing interventions and managing fatigue* *Sense of wanting things proactively suggested*	*Scaffolding of where to find help and information*
		Altering the hearing aid helped *Sense of the NHS aids not being as good as alternatives* *Acknowledgement of limitations of adaptations on NHS* *Desire for the latest technology when possible* *Sense of wanting to know about technological advances – even if not on the NHS* *Cross over hearing aid was useful*	*Information about hearing technology*
		Want for more help *Want for more time to discuss options* *Desire for NF2 specific services*	*Need for more follow ups*
		Desire for more emotional awareness *Potential for more emotional support* *Staff being clear and supportive*	*Awareness of emotional impact*
		Need for specific time and attention to get things right	*Greater awareness of communication needs*

Appendix G: Tables described in main text

Table G1: Age distribution of participants (n=50)

Age Bracket (years)	Frequency	Percentage
18-25	6	12
26-35	5	10
36-45	4	8
46-55	12	24
56-65	11	22
66-75	7	14
76 +	5	10

Table G2: Reported impact of NF2 (n=50)

Impact	Percentage of people reporting
Hearing loss/becoming deafened	92.0
Impaired balance	78.0
Tiredness/ fatigue	52.0
Mental health/self-esteem	44.0
Pain	26.0
Facial palsy	26.0
Reduced vision/loss of sight	24.0
Restricted mobility	24.0
Other	20.0
Other sensory loss	12.0

Table G3: The overlap of themes relating to psychological wellbeing

Quote	Themes related to
"I still get pretty fatigued and tired and if I don't manage things well generally means migraines and confined to bed, followed by a period of lethargy. Which can be stressful for my husband in particular. Which in turn is stressful as you feel you are putting pressure on a loved one."	Burden of physical symptoms: "What a cruel trick our bodies play on us" Social impact and withdrawal: "I often feel left out" Emotional burden: "Thinking about it makes me feel bad" Impact on flexibility and adaptation to cope: "I've been able to adapt, but certain scenarios are still difficult"

"All of the above [listed symptoms of NF2] have had a huge impact on social life - only mixing with one friend at a time, have become introverted meeting new people (or avoid meeting new people), accepting people just think I'm a bit weird."	Burden of physical symptoms: "What a cruel trick our bodies play on us" Social impact and withdrawal: "I often feel left out"
"NF2 has made me have to change career plans, I wanted to join the Royal Navy but the armed forces do not let deaf people or people with brain tumours enlist."	Impact on flexibility and adaptation to cope: "I've been able to adapt, but certain scenarios are still difficult" "Career progression has been significantly impacted" Identity change and loss of confidence: "It feels like sometimes there are no parts of me which are not affected"
"I am much more anxious than I used to be 4 years ago, especially if I need to go into a social situation that is new to me for example a party where I don't know many people or to a sports event where I am participating, to the point where I will at the last moment make an excuse not to go, I get very anxious and have no confidence with people"	Emotional burden: "Thinking about it makes me feel bad" Social impact and withdrawal: "I often feel left out" Identity change and loss of confidence: "It feels like sometimes there are no parts of me which are not affected"

Table G4: Quotes and themes regarding challenge to identity and adaptation

Quote	**Themes related to**
"I cannot where [sic] high or any 'normal' sort of shoe because of my dropped foot.....I hate it, every day!! (not just the lack of shoe choice but just having it!!) My wonky face, catching it in the mirror or photos gets me every time"	Emotional burden: "Thinking about it makes me feel bad" Identity change and loss of confidence: "It feels like sometimes there are no parts of me which are not affected"
"With the support from the Multi-disciplinary team…I have been able to carry on working and I believe manage. I	Burden of physical symptoms: "What a cruel trick our bodies play on us" Emotional burden: "Thinking about it

had to change my career and hours and now work part time…This was a difficult adjustment."	makes me feel bad" "Career progression has been significantly impacted"
"I'm not keen on travelling alone as can get quite agitated that I might miss an announcement, likewise with any appointments. Which I am aware places pressure on others."	Need for adaptation to cope Social impact and withdrawal: "I often feel left out" Identity change and loss of confidence: "It feels like sometimes there are no parts of me which are not affected" Need for adaptation to cope
"Worry about the future - Ability to provide for my family. Concern about whether my children would also be diagnosed with NF2 and the impact on their lives."	Social impact and withdrawal: "I often feel left out" Emotional burden: "Thinking about it makes me feel bad" Identity change and loss of confidence: "It feels like sometimes there are no parts of me which are not affected" "Career progression has been significantly impacted"

Table G5: Examples of hearing aids meeting expectations and the associated themes:

Quote	**Associated themes**
"Since having it installed I have been able to hear much more of what people are saying. One positive element that I was not expecting is that people become much more understanding of my deafness and are far more likely to remember that it is helpful if they speak more clearly when communicating with me as there is the noticeable visual cue fastened to my skull"	Meeting or exceeding expectations: "Hearing aids are the closest thing to a miracle that I can imagine" Social benefits of the hearing aid: "I feel a little less cut off from the world when I use them"
"I have been able to maintain my ability to work. Without a working hearing aid I cannot per[f]orm my day-to-day activities."	Meeting or exceeding expectations: "Hearing aids are the closest thing to a miracle that I can imagine" Hearing aid as vital for functioning: "I would not have been able to function if I did not take it up."

"It exceeded my expectations, when I had it fitted I rediscovered sounds that I forgot were there"	Meeting or exceeding expectations: "Hearing aids are the closest thing to a miracle that I can imagine"
"I often misheard consonants, or simply couldn't hear enough to follow a conversation with several people, or in a noisy location. The hearing aid has increased clarity listening to people, TV, radio...when I take it out and listen to the radio while I fall asleep at night, I'm very aware of it sounded quite muffled in comparison"	Meeting or exceeding expectations: "Hearing aids are the closest thing to a miracle that I can imagine" Social benefits of the hearing aid: "I feel a little less cut off from the world when I use them"

Table G6: Examples of hearing aids not meeting expectations and the associated themes:

Quote	**Associated themes**
"I feel like the device itself could be better. It does seem to struggle unless the environment is already relatively quiet. It's not great at isolating voices, and I don't feel like the sensitivity settings change enough to make a difference. Also, it eats through batteries on an almost daily basis."	Not meeting expectations: "the sound quality was poor and music, especially, was so distorted I couldn't bear it" Practical concerns: "Find it too bulky"
"Could not get the hearing aid to work on a consistent basis"	Not meeting expectations: "the sound quality was poor and music, especially, was so distorted I couldn't bear it" Practical concerns: "Find it too bulky"
"Hearing aid did not make a large improvement and left me feeling disabled."	Not meeting expectations: "the sound quality was poor and music, especially, was so distorted I couldn't bear it" Fear of drawing attention to difference: "I wanted to appear more normal"
"I felt that having a big piece of plastic on the back of my head was just another thing that would mark me out as different"	Fear of drawing attention to difference: "I wanted to appear more normal"

Table G7: Recommendations for improvements

Recommendation for improvement	**Quotes**
Greater awareness of communication needs	*"I wasn't to[o] pleased with the online NF2 consultation as the consultant was wearing a mask, therefore I missed a lot of what he was saying, we all unconsciously lip read"*
	"Some clinics have electronic name signs. Ideally this could be used everywhere" *"During the pandemic video calls were introduced which were very welcome, indeed greater use of VC would be beneficial to everyone. However, why did the NHS introduce a system that did not feature subtitles?"*
Awareness of emotional impact	*"Regular psychological support upon initial diagnosis and also at times of rapid hearing loss (or other deterioration) is absolutely vital."*
	"As much as all the staff are lovely going to the clinic where there are other patients at a later stage than me and seeing what my future holds has been very difficult."
	"They seem to concentrate on the NF2 than us as a person & how it affects our lives."
Need for more follow ups	*"I do feel more time and consideration with specialists would benefit NF2 patients"*
	"I don't feel there was any follow up after I was given the hearing aids"
	"More regular monitoring of hearing loss at times where there is deterioration (as opposed to when hearing seems to be stable)."
Scaffolding of	*"Lip reading classes have been mentioned on a few occasions but*

where to find help and information	*they have not been offered in any tangible form"*
	"I was asked at the Audiology clinic if I'd like to attend something at the hospital for the profoundly deaf where appointments are longer and I would have access to staff further qualified to advise and help in different ways. However I heard nothing more and cannot remember the name of the person who mentioned it or the specific name of the class/clinic"
	"Would be happy to read more about the results of treatments with avastin or similar drugs, or other experimental treatments."
Information about hearing technology	*"Before I was offered an NHS hearing aid I had already privately invested in multiple products…I was grateful to be given an NHS aid but found it was inferior in terms of technology and ease of use"*
	"It would potentially be useful to know about any advancements in technology that could help me with hearing loss"
	"I was told that my current aid is the only one which would be helpful for me but I am not sure if this is because it is the best the NHS is able to offer or if there really is nothing else on the market which would give me a better experience of hearing. I couldn't get an answer about possibly something better if I paid for it"
	"More frequent consideration to switch to newer hearing aid technology as soon as new options become available."

Appendix H: Compliments for the NF2 Team

There were 30 incidences of the theme 'Compliments for the NF2 team' in the data. In their compliments, people described the team as "a great support", "wonderful", "clear", "patient and informative" and "excellent". Some people also expressed a want for the O&SW NF2 Service to be involved sooner in their NF2 diagnosis or in other areas of their care that were not possible due to where the individual was based:

"Would prefer the audiology team at John Radcliffe but it is becoming more challenging to drive there"

"My previous care was managed by [different NF2 team]. I already feel like the team in Oxford have engaged me in more conversations about hearing loss in this short period than the previous 15 years"

Appendix I: Summary of findings to be disseminated to patients

Thank you for your interest in the results of the survey that we recently sent out.

As requested, below we have summarised the main findings:

Wellbeing

People who responded to the survey shared a number of impacts that NF2 has on them. From the data, it was understood that the physical impact of NF2 has a knock-on effect on people's mood, relationships and confidence as well as people needing to adapt their daily lives and career plans to accommodate symptoms.

From this, we have a greater understanding of the psychological impact of NF2 and hope to promote this awareness to allow care to be more psychologically informed.

Hearing aids

1. Most people with hearing aids felt well-informed about their decision to have one and felt they had made decisions based on weighing up information provided.
2. Most people with hearing aids were satisfied with how it had helped their hearing.
3. People who were not satisfied with their hearing aid noted that it had not given them as much sound as they had hoped for, or the sound quality was poor

Cochlear Implants and Auditory Brain Stem Implants

1. We had a total of 4 responses from people with Cochlear Implants and Auditory Brain Stem Implants, so any findings in this area are limited

2. As with the hearing aids, most people with Cochlear Implants and Auditory Brain Stem Implants felt well-informed about their decision to have one and felt they had made decisions based on weighing up information provided.

3. Half of people with these devices felt they were satisfied with them and were able to access sound better.

4. Half of people with these devices did not feel they had helped, or had only helped later when their hearing had decreased further in their other ear.

Service Recommendations

We had many lovely complements for the team and the work they do, as well as really helpful feedback about how we can improve.

Areas to improve were identified as:

1. Being more aware of the emotional impact of NF2

2. Keeping in mind communication needs of our patients and adapting to these (e.g., not wearing a mask on an online consultation or ensuring captions are available)

3. People desire more follow-ups with the team following new interventions or hearing deterioration

4. Information about services that may be useful needs to be easier to access and find- conversations with staff do not always lead to people knowing where and how to access support mentioned.

5. People want to know more information about the latest hearing technology and treatments, even if these may not be available on the NHS.

If you have any questions about these results, or feel you have a suggestion not mentioned, please contact the team.

TDRP Appendices

Appendix A: Target journal (Archives of Sexual Behaviour) instructions for authors, taken from https://www.springer.com/journal/10508/submission-guidelines?gclid=CjwKCAjw3POhBhBQEiwAqTCuBoOTnPoT8uJLRaQluRkt5-9grfLp_1svr34pJc4EgqHHNcZkRairfhoCE1AQAvD_BwE

Manuscript Style

- Type left-justified in 12-point Times New Roman font using 1-inch margins on all sides. Number all pages, except the title page, consecutively with Arabic numerals placed in the upper right-hand corner. In order to facilitate **masked (previously termed "double-blind")** review, leave all identifying information off the manuscript, including the title page and the electronic file name. Appropriate identifying information is attached automatically to the electronic file. Upon initial submission the title page should include only the title of the article.
- An additional title page should be uploaded as a separate submission item and should include the title of the article, author's name (including highest degree received), and author's affiliation. Academic affiliations of all authors should be included. The affiliation should include the department, institution, city, and state (or nation) and should be typed as a numbered footnote to the author's name. The title page should also include the complete mailing address, telephone number, and e-mail address of the one author designated to review proofs.
- An abstract, preferably no longer than 250 words, is to be provided as the second page.

- A list of 4–5 key words is to be provided directly below the abstract. Key words should express the precise content of the manuscript, as they are used for indexing purposes.
- The Acknowledgments section (if any) should be included as part of the **separate title page** to facilitate masked (i.e., **double-blind)** peer review.

Appendix B: The AQ-Short (Hoekstra et al., 2011), used with author permission:

Higher-order factor Social Behavior
Social skills
I prefer to do things with others rather than on my own (1)
I find social situations easy (11)
I would rather go to a library than to a party (13)
I find myself drawn more strongly to people than to things (15)
I find it hard to make new friends (22)
I enjoy social occasions (44)
I enjoy meeting new people (47)
English samples only: New situations make me anxious (46)
Routine
I prefer to do things the same way over and over again (2)
It does not upset my if my daily routine is disturbed (25)
I enjoy doing things spontaneously (34)
New situations make me anxious (46)
Switching
I frequently get strongly absorbed in one thing (4)
I can easily keep track of several different people's conversations (10)
I find it easy to do more than one thing at once (32)
If there is an interruption, I can switch back very quickly (37)
Imagination
Trying to imagine something, I find it easy to create a picture in my mind (3)
Reading a story, I can easily imagine what the characters might look like (8)
I find making up stories easy (14)
Reading a story, I find it difficult to work out the character's intentions (20)
I find it easy to work out what someone is thinking or feeling (36)
I find it difficult to imagine what it would be like to be someone else (42)
I find it difficult to work out people's intentions (45)
I find it easy to play games with children that involve pretending (50)
Factor numbers and patterns
I usually notice car number plates or similar strings of information (6)
I am fascinated by dates (9)
I am fascinated by numbers (19)
I notice patterns in things all the time (23)
I like to collect information about categories of things (41)

Appendix C: The Group-Level Self Definition and Self Investment Scale (Leach et al., 2008), used with author permission:

Items Measuring In-Group Identification

(Group-Level) Self-Investment

Solidarity

1. I feel a bond with [In-group]. (Adapted from Cameron, 2004; Doosje et al., 1998.)
2. I feel solidarity with [In-group].
3. I feel committed to [In-group]. (Doosje et al., 1995)

Satisfaction

4. I am glad to be [In-group]. (Adapted from Cameron, 2004; Doosje et al., 1998; Luhtanen & Crocker, 1992.)
5. I think that [In-group] have a lot to be proud of. (Ellemers, Kortekaas, & Ouwerkerk, 1999.)
6. It is pleasant to be [In-group]. (Doosje et al., 1998.)
7. Being [In-group] gives me a good feeling. (Adapted from Cameron, 2004; Luhtanen & Crocker, 1992.)

Centrality

8. I often think about the fact that I am [In-group]. (Adapted from Cameron, 2004.)
9. The fact that I am [In-group] is an important part of my identity. (Adapted from Luhtanen & Crocker, 1992.)
10. Being [In-group] is an important part of how I see myself. (Adapted from Doosje et al., 1998; Ellemers, Kortekaas, & Ouwerkerk, 1999; Luhtanen & Crocker, 1992.)

(Group-Level) Self-Definition

Individual Self-Stereotyping

11. I have a lot in common with the average [In-group] person. (Adapted from Spears et al., 1997.)
12. I am similar to the average [In-group] person. (Adapted from Doosje et al., 1995; Spears et al., 1997.)

In-Group Homogeneity

13. [In-group] people have a lot in common with each other. (Adapted from Spears et al., 1997.)
14. [In-group] people are very similar to each other. (Adapted from Ellemers, Kortekaas, & Ouwerkerk, 1999; Spears et al., 1997.)

Excluded Items

15. I see myself as [In-group]. (Doosje et al., 1998.)
16. I identify with other [In-group] people. (Doosje et al. (1998); Ellemers, Kortekaas, & Ouwerkerk, 1999.)
17. Being [In-group] just feels natural to me. (Doosje et al., 1998.)
18. I feel (personally) implicated when [In-group] people are criticized. (Adapted from Jackson, 2002.)
19. [In-group] are an important group to me. (Doosje et al., 1998.)
20. I have a lot of respect for [In-group] people. (Ellemers, Kortekaas, & Ouwerkerk, 1999.)

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Appendix D: The Gender Congruence and Life Satisfaction Scale (Jones et al., 2019), used with author permission:

Gender Congruence and Life Satisfaction Scale (GCLS)

Below is a range of statements about how you might feel in relation to your gender, mental well-being, and life satisfaction. Please respond to each statement, thinking about how frequently you have felt like this in the past **6 months**.

Please rate each statement as: NEVER (N), RARELY (R), SOMETIMES (S), OFTEN (O), or ALWAYS (A).

Please note that when talking about ***"gender identity"*** we mean one's internal sense of one's self as a man, a woman, or some other gender.

In the past **6 months**, due to the distress about my gender (i.e., the distress caused as the gender I was assigned at birth does not match with my gender identity):					
1. I have avoided social situations and/or social interactions	N	R	S	O	A
2. I have not gone to school/college/work	N	R	S	O	A
3. I have not been able to have emotional relationships with other people	N	R	S	O	A
4. I have suffered from anxiety	N	R	S	O	A
5. I have not been able to be physically intimate with other people	N	R	S	O	A
6. I have been unable to leave the house	N	R	S	O	A
7. I have found it difficult to make friends	N	R	S	O	A
8. I have thought about cutting or hurting my chest, genitals, and/or surrounding areas	N	R	S	O	A
9. I have felt that life is meaningless	N	R	S	O	A
10. I have not enjoyed life	N	R	S	O	A
11. I have not engaged in leisure activities	N	R	S	O	A
12. I have suffered from low mood	N	R	S	O	A
13. I have thought about hurting myself or taking my own life	N	R	S	O	A

Please rate each statement as: NEVER (N), RARELY (R), SOMETIMES (S), OFTEN (O), or ALWAYS (A).

In the past **6 months**:					
14. I have felt distressed when touching my genitals as they do not match my gender identity	N	R	S	O	A
15. I have felt so distressed about my chest that I have not been able to have a fulfilling life	N	R	S	O	A
16. I have felt comfortable with how others have perceived my gender	N	R	S	O	A
17. I have felt that my body hair conflicts with my gender identity, either because I have it and do not like it or because I would like to have it	N	R	S	O	A
18. I have felt like my chest does not match my gender identity	N	R	S	O	A
19. I have found it distressing that others do not address me according to my gender identity	N	R	S	O	A
20. I have felt satisfied with the pronouns that others use when talking about me	N	R	S	O	A
21. I have felt unhappy about my genitalia since they do not match my gender identity	N	R	S	O	A
22. I have felt comfortable with how other people perceive my gender based on my physical appearance	N	R	S	O	A
23. I have felt that my voice has affected the way other people have perceived my gender identity which has been distressing for me	N	R	S	O	A
24. I have felt that my facial hair conflicts with my gender identity, either because I have it and do not like it or because I would like to have it	N	R	S	O	A
25. I have felt that my genitals do match with my gender identity	N	R	S	O	A
26. I have felt that genital surgery will address the unhappiness I experience in relation to my gender	N	R	S	O	A
☐. Tick here if you have already had genital surgery (unless you feel you need more)					
27. I have been unable to have a fulfilling life because of the distress relating to my genitalia	N	R	S	O	A
28. I have felt extremely distressed when looking at my chest	N	R	S	O	A
29. I have felt extremely distressed when looking at my genitals	N	R	S	O	A
30. I have felt satisfied with my chest	N	R	S	O	A

Please rate each statement as: NEVER (N), RARELY (R), SOMETIMES (S), OFTEN (O), or ALWAYS (A).

Next, we would like to know how satisfied you have been with your life for the last **6 months**:					
31. I have felt satisfied at school/college/work	N	R	S	O	A
32. I have felt satisfied with my emotional relationship(s)	N	R	S	O	A
33. I have felt satisfied with my sex life	N	R	S	O	A
34. I have felt satisfied in my leisure activities and hobbies	N	R	S	O	A
35. I have not felt satisfied with my friends	N	R	S	O	A
36. I have felt satisfied with the support I have received from other significant people	N	R	S	O	A
37. I have not felt satisfied with my health	N	R	S	O	A
38. I have felt satisfied with life in general	N	R	S	O	A

Appendix E: Social media recruitment

Facebook	Instagram	Known Snowball Sampling Shares
Trans Masculine Support and Advice UK	Pink Therapy	Birmingham Pride WhatsApp Group
Banbury LGBT+	The Oxford Psych	GIRES
TransActual		

Appendix F: Tests of normality

Descriptives

Descriptive Statistics

AQ_percentile_split		N	Minimum	Maximum	Mean	Std. Deviation	Skewness		Kurtosis	
		Statistic	Statistic	Statistic	Statistic	Statistic	Statistic	Std. Error	Statistic	Std. Error
Low	Social_Identity_Self_investment	62	20.00	50.00	40.1452	6.83013	-.837	.304	.235	.599
	Social_Identity_Self_definition	62	4.00	20.00	12.1774	3.44286	.156	.304	-.073	.599
	Valid N (listwise)	62								
High	Social_Identity_Self_investment	66	24.00	50.00	39.6212	7.64957	-.593	.295	-.787	.582
	Social_Identity_Self_definition	66	4.00	20.00	12.6667	3.19294	.229	.295	.480	.582
	Valid N (listwise)	66								

Descriptives

Descriptive Statistics

AQ_percentile_split		N	Minimum	Maximum	Mean	Std. Deviation	Skewness		Kurtosis	
		Statistic	Statistic	Statistic	Statistic	Statistic	Statistic	Std. Error	Statistic	Std. Error
Low	GCLS_Cluster1_Gender_Congruence	57	1.35	3.82	2.5408	.60679	.219	.316	-.344	.623
	GCLS_Cluster2_wellbeing_life_satisfaction	59	1.33	3.86	2.5052	.64375	.259	.311	-.731	.613
	Valid N (listwise)	56								
High	GCLS_Cluster1_Gender_Congruence	63	1.41	3.65	2.3819	.59426	.469	.302	-.762	.595
	GCLS_Cluster2_wellbeing_life_satisfaction	65	1.19	3.90	2.1853	.54393	.591	.297	.703	.586
	Valid N (listwise)	63								

Descriptive Statistics

AQ_percentile_split		N Statistic	Minimum Statistic	Maximum Statistic	Mean Statistic	Std. Deviation Statistic	Skewness Statistic	Skewness Std. Error	Kurtosis Statistic	Kurtosis Std. Error
Low	Social_Identitiy_Solidarity	62	8.00	15.00	13.4032	1.94565	-1.132	.304	.457	.599
	Social_Identitiy_Satisfaction	62	7.00	20.00	14.5484	3.59269	-.546	.304	-.579	.599
	Social_Identity_Centrality	62	3.00	15.00	12.1935	2.83328	-1.428	.304	2.147	.599
	Social_Identity_Self_stereotyping	62	2.00	10.00	6.5000	1.95649	-.278	.304	-.283	.599
	Social_Identity_Homogeneity	62	2.00	10.00	5.6774	1.86226	.270	.304	-.299	.599
	Valid N (listwise)	62								
High	Social_Identitiy_Solidarity	66	3.00	15.00	13.0000	2.56605	-1.825	.295	4.102	.582
	Social_Identitiy_Satisfaction	66	5.00	20.00	14.2727	3.97123	-.366	.295	-.769	.582
	Social_Identity_Centrality	66	6.00	15.00	12.3485	2.40827	-.793	.295	-.073	.582
	Social_Identity_Self_stereotyping	66	2.00	10.00	6.6970	1.83948	-.424	.295	.427	.582
	Social_Identity_Homogeneity	66	2.00	10.00	5.9697	1.78859	.313	.295	.267	.582
	Valid N (listwise)	66								

Descriptive Statistics

AQ_percentile_split		N Statistic	Minimum Statistic	Maximum Statistic	Mean Statistic	Std. Deviation Statistic	Skewness Statistic	Skewness Std. Error	Kurtosis Statistic	Kurtosis Std. Error
Low	GCLS_genitalia	57	1.00	4.00	2.6491	.70054	-.429	.316	-.520	.623
	GCLS_chest	60	1.00	4.00	2.4292	1.20916	.106	.309	-1.679	.608
	GCLS_2nd_sex_characteristics	60	1.00	4.00	2.4333	.96901	.094	.309	-1.203	.608
	GCLS_social_gender_role_recog	60	1.25	4.00	2.7167	.77222	.158	.309	-.874	.608
	GCLS_physical_emotional_intimacy	59	1.00	4.00	2.5551	.86983	-.169	.311	-1.089	.613
	GCLS_psycholo_funct	61	1.00	4.00	2.3262	.85380	.385	.306	-.839	.604
	GCLS_life_satisfaction	59	1.57	4.00	2.7094	.52854	.062	.311	-.355	.613
	Valid N (listwise)	56								
High	GCLS_genitalia	63	1.17	3.83	2.4524	.71368	.015	.302	-.971	.595
	GCLS_chest	65	1.00	4.00	2.2000	1.12777	.394	.297	-1.349	.586
	GCLS_2nd_sex_characteristics	65	1.00	4.00	2.2974	.94487	.215	.297	-1.127	.586
	GCLS_social_gender_role_recog	65	1.25	4.00	2.5654	.77595	.450	.297	-.965	.586
	GCLS_physical_emotional_intimacy	65	1.00	4.00	2.1692	.73568	.647	.297	-.367	.586
	GCLS_psycholo_funct	65	1.00	4.00	2.0077	.70496	.806	.297	.629	.586
	GCLS_life_satisfaction	65	1.43	3.86	2.4484	.51190	.258	.297	-.017	.586
	Valid N (listwise)	63								