A thesis submitted in partial fulfilment of the requirements of the degree of Doctor of Clinical Psychology (DClinPsych)

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Systematic Review of the Literature (SRL)	6,492
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Abstract (SRL)

Systematic Review of the Literature: What factors are associated with engagement in and adherence to psychological therapy for adults with Non-Epileptic Attack Disorder?

Purpose. The purpose of this review was to collate and synthesise the literature on the factors associated with engagement and adherence to psychological therapy as a treatment for adults with Non-Epileptic Attack Disorder (NEAD).

Methods. Five electronic databases were systematically searched (PsycInfo, PubMed, CINAHL, MEDLINE, and EMBASE). The quality of these was assessed using the Mixed Methods Appraisal Tool.

Results. The search was performed across five databases on the 22nd February 2022, and returned 2,150 papers. Thirteen papers were included in the review, which were of varying quality. Three of the papers were qualitative and 10 were quantitative. The papers reported several different factors associated with engagement and adherence, which spanned across the five dimensions of the World Health Organisation Multidimensional Adherence Model. Demographic characteristics associated with adherence include older age, having a carer, taking anti-epileptic medications, and being unemployed. Reasons people gave for not engaging with or adhering to psychological therapy included logistical barriers, not feeling ready, and not agreeing with the diagnosis of NEAD. A single motivational interviewing session, a brief NEAD educational session, and offering telepsychology appointments was found to significantly increase adherence.

Conclusion. The factors associated with engagement and adherence to psychological therapy in NEAD are multifaceted and are not all patient related factors, despite most research being in this area. Future research should involve trialling interventions while controlling for the time spent with the patient.

Abstract (SIP)

Service Improvement Project: Investigating the decision-making process regarding surgery for parents of children with craniosynostosis at the Oxford Craniofacial Unit.

Objective. This Service Improvement Project explored the current decision-making process regarding corrective surgery for Craniosynostosis; a decision made by parents/carers on behalf of their children. The study examines what information parents use to make this decision, as well as exploring what is being done well, and what could be improved within the service.

Design. Online qualitative surveys were used to collect data.

Setting. The study was conducted at one of the five highly specialised Craniofacial centres in the UK.

Participants. Eighteen parents of children with Craniosynostosis and ten MDT members participated in the project.

Main Outcome Measure(s). Qualitative surveys were analysed using Thematic Analysis. The parent and MDT surveys were analysed separately. Analysis was guided by the Theory of Reasoned Action and Theory of Planned Behaviour.

Results. The analysis showed that the MDT are promoting shared decision making across the team. Parents base their decision on a number of factors, including their risk tolerance, values around appearance, and the opinions of others (e.g. society and family/friends). The parents discussed the challenges of the decision and the associated difficult emotions, but praised the support offered by the Craniofacial Team. Suggested improvements to the process are detailed.

Conclusions. The decision about corrective surgery is an emotive experience for parents, however this is a process parents feel supported with by the Oxford Craniofacial Team. Suggestions to improve this process for parents even further are listed.

Abstract (TDRP)

Theory Driven Research Project: Exploring the Impact of Perceived Social Support as a Moderator of the Relationship Between Perceived Public Stigma and Negative Outcomes in Bell's Palsy

Objective. The main aims of this study were to explore the differences in perceived social support between people with Bell's Palsy (BP) and the general population, and to test for a potential association between perceived stigma and psychological distress in the BP population. Perceived social support was tested as a potential moderator of this relationship.

Design. This study employed a cross-sectional survey design.

Participants. Ninety-four people with BP and ninety-one controls without BP participated in this study.

Main Outcome Measure(s). Perceived Stigma Questionnaire, Multidimensional Scale of Perceived Social Support, Patient Health Questionnaire-8 (depression measure), and Generalised Anxiety Disorder Assessment-7.

Results. People with BP reported significantly lower levels of perceived social support and higher levels of depression and anxiety compared to people in the general population. Perceived stigma was significantly associated with (and predicted) negative psychological outcomes, but perceived social support did not moderate this relationship.

Conclusions. Limitations of the study are discussed. Future research should explore other potential moderators of the relationship between stigma and psychological outcomes.

Systematic Review of the Literature

What factors are associated with engagement in and adherence to psychological therapy for adults with Non-Epileptic Attack Disorder?

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Proposed Journal

Epilepsy and Behaviour is an international journal that welcomes papers about the behavioural aspects of seizures and epilepsy. Previous publications include a number of the articles selected for this systematic review. Author submission guidelines are found in Appendix A.

Acknowledgements

This work was supported by the Oxford Institute for Clinical Psychology Training and Research. A second trainee on the course, Jerica Radez (JR), was involved in the project as a collaborator to perform reliability checks.

Funding

No funding was sought for this project.

Declaration of Interest

There are no conflicts of interest.

Abstract

Purpose. The purpose of this review was to collate and synthesise the literature on the factors associated with engagement and adherence to psychological therapy as a treatment for adults with Non-Epileptic Attack Disorder (NEAD).

Methods. Five electronic databases were systematically searched (PsycInfo, PubMed, CINAHL, MEDLINE, and EMBASE). The quality of these was assessed using the Mixed Methods Appraisal Tool.

Results. The search was performed across five databases on the 22nd February 2022, and returned 2,150 papers. Thirteen papers were included in the review, which were of varying quality. Three of the papers were qualitative and 10 were quantitative. The papers reported several different factors associated with engagement and adherence, which spanned across the five dimensions of the World Health Organisation Multidimensional Adherence Model. Demographic characteristics associated with adherence include older age, having a carer, taking anti-epileptic medications, and being unemployed. Reasons people gave for not engaging with or adhering to psychological therapy included logistical barriers, not feeling ready, and not agreeing with the diagnosis of NEAD. A single motivational interviewing session, a brief NEAD educational session, and offering telepsychology appointments was found to significantly increase adherence.

Conclusion. The factors associated with engagement and adherence to psychological therapy in NEAD are multifaceted and are not all patient related factors, despite most research being in this area. Future research should involve trialling interventions while controlling for the time spent with the patient.

1. Introduction

1.1 Non-epileptic Attack Disorder (NEAD)

Non-epileptic attacks (also referred to as functional, dissociative, or psychogenic seizures) are a symptom of Functional Neurological Disorder (FND) in the Diagnostic and Statistical Manual, 5th edition (1). Non-epileptic attacks may on the surface resemble epileptic seizures, however they are not caused by abnormal neurological electrical activity but are considered to be linked to a dysfunction in emotional processing and social distress (2). NEAD often co-occurs with epilepsy (3). Risk factors for developing NEAD include previous significant trauma, current stressful life events, and exposure to other neurological conditions (4).

Around one in five individuals presenting to an epilepsy clinic will be diagnosed with NEAD (5). It more commonly occurs in women (6). NEAD is diagnosed by ruling out other potential causes, such as epilepsy and syncope, a process which can take several years (7). The gold standard of achieving a diagnosis is capturing an attack during EEG monitoring and finding no changes to neurological electrical activity before, during or after the seizure (8). Despite their differing causes, NEAD has been shown to be just as disabling as epilepsy, due to, for example, its impact on family functioning and ability to work (9).

1.2 Current Treatment for NEAD

A number of psychological interventions have been shown to be effective in reducing or stopping patients' attacks, including Cognitive Behavioural Therapy (CBT), psychoeducation, and mindfulness (10, 11). There are currently no NICE guidelines about treatment recommendations for FND. Psychological intervention of some sort is the treatment of choice for physicians diagnosing NEAD (12). This is an emerging field of research, and evidence for the use of one psychological intervention over another is limited (13). As evidence emerges, it is important to understand the wider context of the implementation of these interventions (14).

Despite the efficacy of psychological interventions for NEAD, the uptake of and adherence to these is poor (15). Consequently, people with NEAD are often known as a *"difficult to engage"* population (16, p. 225). Non-adherence in NEAD is associated with higher seizure frequency, worse quality of life, and increased attendance to the emergency department (17). Indeed, it has been stated by the World Health Organisation that *"increasing the effectiveness of adherence interventions may have a far greater impact on the health of the population than any improvement in specific medical treatments"* (18, p. XIII).

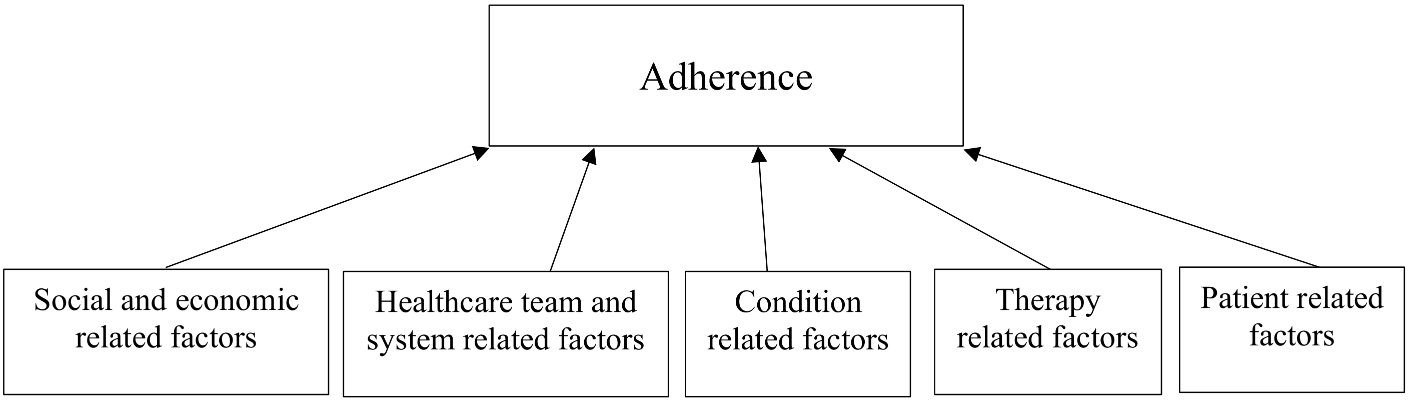
1.3 Conceptualising Engagement and Adherence

In this review, the terms 'engagement' and 'adherence' are used as factual, non-judgemental, and non-blaming terms. For the purposes of this review, engagement is defined as beginning treatment recommended by a healthcare professional. Adherence is defined by the World Health Organisation as *"the extent to which a person's behaviour – taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed recommendations from a health care provider"* (19, p. 3). In this review, the term adherence is used to describe continuing to attend a psychological intervention, as recommended by a healthcare provider.

Previously, studying adherence meant exploring problems with patient compliance, however, a more systemic approach is now recommended (19). Adherence is now seen as a complex, dynamic, and multifaceted concept for which patients, health-care providers, and the wider system are responsible (15). The World Health Organization's Multidimensional Adherence Model (WHOMAM, Figure 1.1) is a comprehensive, systemic model which outlines the factors which influence adherence (19). It acknowledges that most research into adherence focuses on medication, but the model can be used to describe adherence to any health-related behaviour, including psychological therapy. The model proposes five interacting dimensions which influence adherence: 1). '*Social and economic related factors'* which includes support networks, socio-economic status, and unemployment; 2). '*Healthcare team and system related factors'* which can include the therapeutic relationship and knowledge of healthcare providers; 3). '*Condition related factors*' which are the condition-specific demands placed on the patient, such as level of disability and severity of symptoms; 4). '*Therapy related factors'* which encompasses any logistical barriers to accessing treatment; and 5). '*Patient related factors'* which comprises of illness beliefs, motivation to engage, and any demographic characteristics. The latter is the most well researched dimension of the model. The model has been used to research adherence to recommended treatment in a number of physical health conditions, such as heart failure (20). The World Health Organisation highlight that adherence must be understood for each specific population, as it may be affected by distinct illness-related demands. Given the fact that NEAD sits at a unique crossing between neurology and psychiatry, it is important to explore the factors that affect adherence in this population specifically.

Figure 1.1

World Health Organisation Multidimensional Adherence Model



Changed from 'Medication Adherence' in the original model, reproduced with permission from the World Health Organisation (Appendix B)

1.4 Review Aims

Although a recent systematic review investigated the efficacy of psychological treatment in this population (21), there has not been a review investigating engagement and adherence to therapy. This review aims to collate studies which explore the factors that are associated with engagement and adherence to psychological intervention as a treatment for adults with NEAD. This includes studies of interventions aiming to increase engagement and adherence. The results gained from the review will be put into context using the WHOMAM. Once we have a greater understanding of what is preventing this population from engaging in and adhering to psychological interventions, methods to address this can be explored further.

2. Method

The protocol was registered on PROSPERO (ID: CRD42021270402; available at https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=270402).

2.1 Eligibility Criteria

2.1.1 Inclusion Criteria

- Studies investigating the factors associated with engagement and adherence to psychological therapy as a treatment for NEAD in adults (18 years and over)
- Studies aimed at increasing engagement or adherence to psychological therapy for adults with NEAD
- Participants are adults with NEAD or healthcare professionals working with adults with NEAD
- Studies using qualitative, quantitative, and mixed-method methodologies
- Empirical papers, including case studies
- Grey literature

2.1.2 Exclusion Criteria

- Studies involving people 17 years old or younger
- Papers not written in English
- Papers exploring engagement or adherence to psychological therapy in epilepsy
- Literature reviews, editorial letters, and theoretical papers
- Studies investigating access (defined as offering psychological therapy as a mental health treatment) to psychological therapy for NEAD
- Studies investigating engagement or adherence to psychological therapy for patients with FND as an umbrella diagnosis

2.2 Search Strategy

The following databases were searched on 22nd February 2022: PsycInfo, PubMed, CINAHL, MEDLINE, and EMBASE. Search terms included ‘NEAD’ and other diagnostic labels, combined with ‘engagement’ or ‘adherence’ or other related terms, and ‘psychotherapy’ or other related terms (Appendix C). The search was re-run on 17th April 2023, with no additional papers found.

Grey literature was included in this search. The first author contacted authors of conference abstracts to ask for full details. Forwards citation searching using Google Scholar was conducted on 17th May 2022, examining papers which had referenced included studies (22). Backwards citation searching was conducted by a manual search of the reference lists of all articles meeting the inclusion criteria.

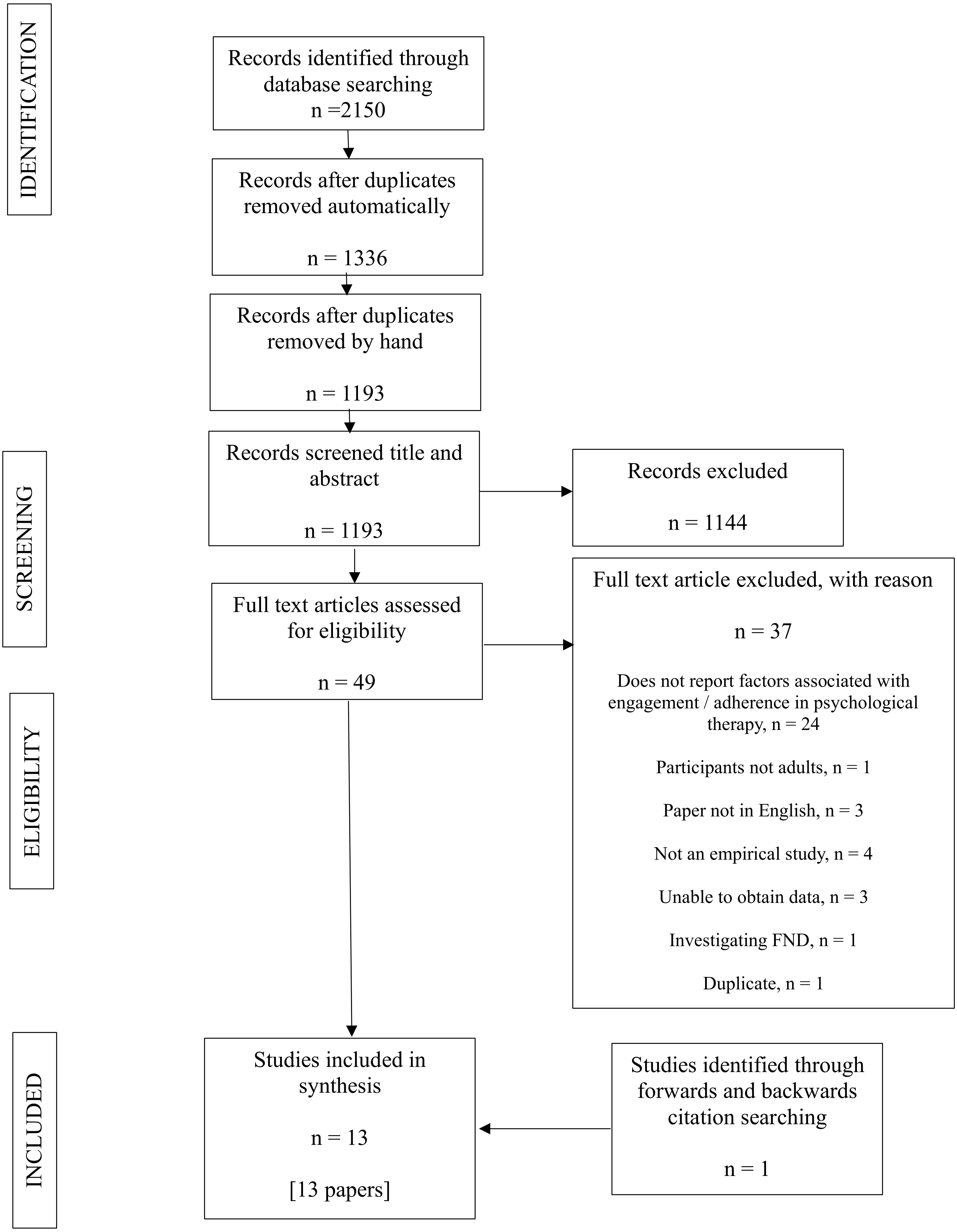
2.3 Study Selection

A Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) flowchart is presented in Figure 1.2 (23). After removing duplicates, 1193 papers were screened for eligibility, according to the paper’s title and abstract. Papers that did not meet the criteria were excluded. A second reviewer (JR) screened 20% of the papers. A strong level of agreement was reached between the two reviewers, $k = 0.86$. Discrepancies in the inclusion of a paper was discussed between the reviewers. All papers included by both reviewers advanced to the next stage.

Forty-nine papers were reviewed at the full-text stage. Thirty-seven failed to meet the criteria, leaving 12 that met inclusion criteria. One paper was identified during forwards citation searching. Therefore, thirteen papers were included in the review. A second reviewer (JR) independently reviewed 25% of the full texts. There was perfect agreement between the two reviewers, $k = 1$.

Figure 1.2

Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) Diagram



2.4 Data Extraction and Quality Assessment

A data extraction table was developed, to extract relevant information in a systematic way. The first reviewer extracted data from all 13 studies. The second reviewer extracted data from 50% of the studies. The results were compared, and any differences were discussed.

The methodological quality of the included studies was assessed using the Mixed Method Appraisal Tool (MMAT, Appendix D), which covers qualitative, quantitative, and mixed-method study designs (24). The studies were first screened with two questions assessing clarity of the study's aims and methods. If the answers were 'No' then the study did not progress to the quality assessment phase. The quality assessment questions differed depending on the study design. Questions are answered with a 'Yes', 'No', or 'Can't tell' response. The second reviewer independently reviewed 50% of the studies and a strong agreement was found between the reviewers' quality ratings, $k = 0.87$.

2.5 Data Synthesis

A narrative synthesis was conducted, following the Economic and Social Research Council (ESRC) guidance (25). This approach was chosen due to the predominantly descriptive nature of the results. Additionally, this synthesis technique is noted to be particularly useful when exploring the factors impacting the implementation of a treatment (25). The ESRC guidance recommends three main stages of analysis: (1) Developing a preliminary synthesis of findings of included studies, (2) Exploring relationships within the data, and (3) Assessing the robustness of the synthesis. Different techniques are suggested for use at each stage, with the guidance recommending researchers choose which technique is best suited to their research question. Narrative synthesis has been effectively used in a number of other studies exploring engagement in psychological therapy (e.g. 26).

The aim of developing a preliminary synthesis is to familiarise oneself with the results of the studies, and to begin to identify the factors which impact the implementation of the intervention. To achieve this, a textual description of each of the studies was produced. These were written in a systematic way, including the same details for each study.

Two techniques were used to explore the relationships within the data. The first was 'groupings and clusters' which involves organising the included studies into smaller groups, based on the research question. This can help to see patterns within and across groups. The second was 'ideas webbing' which involves creating a visual picture of possible relationships between the results of different studies. The WHOMAM was used as a framework. Each barrier/facilitator to engagement/adherence was grouped by which overarching dimension they related to. Within this, similar findings from studies were grouped within themes.

To ensure the robustness of the synthesis, a sensitivity analysis was performed. 'Low' quality studies (not passing screening or ≥2 answers of 'No' in the MMAT) were temporarily excluded from the analysis to determine whether the results were the same. Additionally, the primary author used the recommendations set out by Busse et al. to critically reflect on the process (27). This includes discussing the quality, validity and generalisability of the evidence used, as well as the limitations of the synthesis.

3.0 Results

3.1 Study Characteristics

The review included 13 studies. Three of these were qualitative (28–30) and ten were quantitative (14, 15, 17, 31–37). One of these was an unpublished thesis (31). The studies fell into two categories. The first (Table 1.1) reported on the factors associated with engagement and adherence. This was either done by exploring which baseline characteristics were associated with non-engagement and non-adherence (14, 17, 32, 33, 36), or by reporting the reasons that patients gave for not engaging with or adhering to therapy (15, 28–32). The second category (Table 1.2) assessed the impact of interventions designed to improve engagement and adherence (14, 34–37).

The age of participants ranged from 18-84. All studies had more female participants. Eight were conducted in the USA (14, 17, 28, 31, 34–37), four in the UK (29, 30, 32, 33) and one in France (15). They looked at engagement and adherence to various psychological therapy treatments for NEAD, including CBT, psychoeducation, and mindfulness.

The majority looked at engagement in and adherence to individual therapy; only one looked at group therapy (32). Most participants had their NEAD diagnosed by an Epileptologist or Neurologist, after habitual events were captured on video-EEG with no associated abnormal electrical discharges. Half of the studies excluded people if they had co-morbid epilepsy (28, 30, 31, 34, 35). The measurement of engagement and adherence varied, ranging from having a therapy session booked, to patients still having ongoing psychological input at the time of the study. Only two of the intervention studies designed to improve engagement and adherence were randomised controlled trials (34, 37).

Table 1.1

Papers which investigate the factors associated with engagement and adherence

Citation	**Study design**	**Number of participants**	**Age**	**Country**	**Females (%)**	**Type of therapy**	**Definition of engagement or adherence**	**Summary of relevant findings**	**Quality assessment**
Andrini et al. (2022)	Qualitative interview	1	44	USA	100%	CBT delivered by CBT therapist	Adherence defined as attending three CBT sessions	Reasons given for non-adherence: • Not feeling validated by the therapist. • Lack of rationale for why CBT would be helpful • Therapist not knowledgeable enough about NEAD • Did not think CBT would have lasting effects	4 *
Baslet & Prensky (2013)	Quantitative, cross-sectional	85	mean = 39.16 SD = 13.68	USA	21.2%	No information on approach	Adherence defined as attending at least five therapy sessions with therapist within the NEAD clinic, or attending therapy for 2 months elsewhere	Factors predicting non-adherence: • Not being married (or not having a live in partner) • Reporting cognitive complaints Factors associated with non-adherence: • Exposure to anti-epileptic drugs	2 *

Tilahun et al. (2022)	Quantitative, cross-sectional	257	mean = 36.9 SD = 14.3	USA	79%	12-session CBTip (Cognitive Behavioural Therapy-informed psychological intervention) at an outpatient PNES clinic, either delivered in person or via telemedicine	For each of the 12 sessions, visit status was classified as attended, cancelled, no-show, or left without seen	Factors associated with adherence: • Older age significantly associated • Higher median income (by ZIP code)	5*
Fettig et al. (2020)	Quantitative, cross-sectional	108	mean = 34 SD = 10.8	France	72.2%	No information on approach	Dis-engaging at any point during psychological therapy	Reasons given for non-adherence: • Lack of interest • Feeling better • Lack of confidence in mental health professionals	3 *
Flom (1997)	Quantitative, cross-sectional	53	range = 18-79 [a]	USA	85% [a]	Varied in approach, healthcare profession and frequency	No info	Reasons given for non-adherence: • Did not agree with the recommendation • Financial reasons • Disagreed with the form of therapy • Felt better	2 *

Jones et al. (2021) [b]	Quantitative, cross-sectional	64	Engaged range = 22 – 72 Disengaged range = 18 - 66	UK	Engaged = 72% Disengaged = 84.6 %	Online CBT group consisting of 6 sessions of 90 minutes	Adherence defined as attending the first session of the CBT group	Factor associated with non-adherence: • Younger age	3 *
	Quantitative, cross-sectional	18	no info		no info			Reasons given for non-adherence: • Unable to attend due to the time or regularity of the sessions • Did not like the group / online format • Tech issues	

Read et al. (2020)	Qualitative interview	30	18-80	UK	70%	NEAD specific CBT intervention consisting of 12 one hour-long sessions, delivered by a CBT therapist	No info	Themes and sub-themes around reasons given for non-adherence: • Worried about 'dragging up the past' • Too big a commitment in terms of travel • Could not see the value of CBT, just wanted to talk • Did not feel ready to discuss trauma • Had not formed an alliance with the therapist • Therapy was too distressing • Therapy increased seizure frequency • Decided to engage as it offered hope in relation to treatment	5 *
Stone et al. (2020)	Quantitative, cross-sectional	698	range = 18-84	UK	73.8%	Psychiatry appointment prior to NEAD specific CBT intervention, which consisted of 12 one hour-long sessions, delivered by a CBT therapist	Adherence defined as attending initial Psychiatry appointment	Factor associated with engagement: • Having a carer	4 *

Tolchin et al. (2019)	Quantitative, cross-sectional	87	range = 18-82 [a]	USA	85% [a]	Licensed social worker therapist at the hospital delivered treatment consisting of 12 weekly hour-long mindfulness-based therapy sessions, *or* treatment was provided by a local therapist if the participant already had an existing therapeutic relationship or lived >1–2 hours from the hospital.	Adherence defined as attending at least eight psychotherapy sessions over 16 weeks	Factors associated with non-adherence: • Unemployed and being on disability at the time of diagnosis • Lower occupational status Factors predictive of non-adherence: • Self-identified racial or ethnic minority status • History of childhood abuse	5 *

Wyatt et al. (2014)	Qualitative interviews	6	range = 29-55	UK	83%	Outpatient session with a psychologist within an adult neuropsychology NHS service	Adherence defined as attending at least one outpatient session with a Psychologist	Reasons given for non-engagement/adherence: • Previous experiences of feeling disbelieved by healthcare professionals discouraged from engaging • Worry about the connotations of seeing a mental health professional • Confused by the referral in the context of ongoing neurological investigations • Therapy is powerful and helpful, but *"hazardous in nature"* Factors which promoted engagement/adherence: • Encouragement from family members • Positive ethos of the whole neuropsychiatry team	4 *

Note. For the reporting of the quality assessment, one answer of 'yes' equates to one star (*) on the MMAT
Baslet & Prensky (2013) and Tilahun et al. (2022) appears in both tables
Age information given is what was presented in original papers
Associated characteristics were calculated by statistically comparing demographics of the people who engaged/adhered compared to those who did not
Predictive characteristics were calculated with cross-sectional logistic regression analysis
[a] Demographics taken from main study
[b] Jones et al (2022) study compromises of two sections, both listed in the table

Table 1.2

Papers which investigate interventions to increase engagement or adherence

Citation	Number of participants	Age	Country	Females	Intervention to improve engagement / adherence	Control group	Allocation to group	Definition of engagement / adherence	Outcome	Quality assessment
Baslet & Prensky (2013)	85	mean = 39.16 SD = 13.68	USA	21.2%	Integrated treatment (psychiatry and psychology in the same clinic)	Divided treatment (psychological therapy elsewhere)	Dependent on geographical limitations	Adherence defined as attending at least five therapy sessions with therapist within the NEAD clinic, or attending therapy for 2 months elsewhere	A higher percentage adhered in the integrated group, but this was not significant	2 *
Thompson et al. (2005)	50	no info	USA	no info	Brief educational intervention following a modified version of the Shen et al. (1990) approach²	n.a.	All participants in intervention group	Adherence defined as attending at least one Psychotherapy session	No statistical analysis performed	Did not pass screening

Tilahun et al. (2022)	257	mean = 36.9 SD = 14.3	USA	79%	Online Telepsychology CBT appointments	In person CBT sessions	Participants choose whether they receive in person psychology or telepsychology	For each of the 12 sessions, visit status was classified as attended, cancelled, no-show, or left without seen	Telepsychology appointments were significantly more likely to be attended	5 *
Tolchin et al. (2019)	60	Intervention group mean = 40.7 (SD = 14.3) Control group mean = 39.6 (SD = 16.8)	USA	Intervention group = 84% Control group = 79%	Single 30-minute motivational interviewing session prior to starting weekly psychological intervention	Psychological intervention alone	Randomised	Adherence defined as attending at least eight psychotherapy sessions over 16 weeks	Participants who received the motivational interviewing session were significantly more like to adhere to the psychological intervention	5 *
Thompson et al. (2012)	19	mean = 33	USA	60%	Brief educational intervention following a modified version of the Shen et al. (1990) approach[2]	Neurologist informs the patient that the seizures are non-epileptic and suggests seeking mental health care	Randomised	Adherence defined as having made an appointment with a mental health professional	Participants who received the intervention were significantly more likely to have made or attended a therapy appointment	4 *

Note. For the reporting of the quality assessment, one answer of ‘yes’ equates to one star (*) on the MMAT

[2] Please see results below for full description of approach

3.2 Quality Assessment

In the MMAT, five questions were asked of each study, therefore the highest rating possible is 5*, and the lowest is 0*. Two studies were rated as low quality, scoring 0-2* on the MMAT (14, 31). One did not pass the screening stage as the methods were not appropriate to answer the research question, it is therefore assumed to be a low quality study (35). Six were rated as medium quality, scoring 3-4* on the MMAT (15, 28, 30, 32–34). Four studies were rated as high quality, scoring the maximum of 5* on the MMAT (17, 29, 36, 37). Table 1.3 presents the outcome of the quality assessment (Y = Yes, N = No, CT = Can't tell). There were no mixed-method studies.

A weak aspect of several of the studies was using a non-standardised questionnaire with pre-determined options to assess why people had not engaged or adhered to psychological therapy. This limited the answers that participants were able to give. Additionally, the risk of non-response bias in most studies was high, as they had a poor response rate. All studies were included in the synthesis, irrespective of quality.

Table 1.3

Quality Assessment of Qualitative Studies

	Andrini et al. (2022)	Read et al. (2020)	Wyatt et al. (2014)
S1. Are there clear research questions?	Y	Y	Y
S2. Do the collected data allow to address the research questions?	Y	Y	Y
1.1. Is the qualitative approach appropriate to answer the research question?	Y	Y	Y
1.2. Are the qualitative data collection methods adequate to address the research question?	Y	Y	CT
1.3. Are the findings adequately derived from the data?	N	Y	Y
1.4. Is the interpretation of results sufficiently substantiated by data?	Y	Y	Y
1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?	Y	Y	Y
Total quality assessment score	4*	5*	4*

Quality Assessment of Quantitative Randomised Controlled Trials

	Tolchin et al. (2019)	Thompson et al. (2013)
S1. Are there clear research questions?	Y	Y
S2. Do the collected data allow to address the research questions?	Y	Y
2.1. Is randomization appropriately performed?	Y	Y
2.2. Are the groups comparable at baseline?	Y	CT
2.3. Are there complete outcome data?	Y	Y
2.4. Are outcome assessors blinded to the intervention provided?	Y	Y
2.5 Did the participants adhere to the assigned intervention?	Y	Y
Total quality assessment score	5*	4*

Quality Assessment of Quantitative Non-randomised Studies

	Baslet & Prensky (2013)	Tilahun et al. (2022)
S1. Are there clear research questions?	Y	Y
S2. Do the collected data allow to address the research questions?	Y	Y
3.1. Are the participants representative of the target population?	CT	Y
3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?	CT	Y
3.3. Are there complete outcome data?	Y	Y
3.4. Are the confounders accounted for in the design and analysis?	N	Y
3.5. During the study period, is the intervention administered (or exposure occurred) as intended?	Y	Y
Total quality assessment score	2*	5*

Quality Assessment for Quantitative Descriptive Studies

	Fettig et al. (2020)	Flom (1997)	Jones et al. (2021)	Stone et al. (2020)	Thompson et al. (2005)	Tolchin et al. (2019)
S1. Are there clear research questions?	Y	Y	Y	Y	N	Y
S2. Do the collected data allow to address the research questions?	Y	Y	Y	Y	CT	Y
4.1. Is the sampling strategy relevant to address the research question?	Y	Y	Y	Y	n.a.	Y
4.2. Is the sample representative of the target population?	Y	CT	Y	Y	n.a.	Y
4.3 Are the measurements appropriate?	N	N	N	Y	n.a.	Y
4.4 Is the risk of non-response bias low?	N	N	N	N	n.a.	Y
4.5. Is the statistical analysis appropriate to answer the research question?	Y	Y	Y	Y	n.a.	Y
Total quality assessment score	3*	2*	3*	4*	n.a.	5*

Key: Y = Yes, N = No, CT = Can’t tell

3.3 Themes

Guided by the WHOMAM, themes were identified within each of the dimensions of adherence (Table 1.4).

Table 1.4

Synthesis Themes

WHOMAM Dimension	Theme
Social and economic related factors	Social support network Economic factors
Healthcare team and system related factors	Present therapeutic relationship Previous experiences with the healthcare system
Condition related factors	Mixed messages Effect on seizures
Therapy related factors	Practical barriers Interventions to address therapy related factors
Patient related factors	Characteristics Disagree with approach or recommendations Not feeling ready Interventions to address patient related factors

3.3.1 Social and Economic Related Factors

3.3.1.1 Social Support Network.

Evidence for the impact of having social support comes from both qualitative and quantitative studies. Being married, or having a cohabiting partner, was associated with higher rates of initial engagement (14). In this study marital status was found to be predictive of adherence, defined as attending at least five sessions of therapy within the service, or remaining active in therapy for 3 months elsewhere. It is worth noting a possible selection bias in this study though, as it only included participants who voluntarily agreed to neuropsychiatric follow up after diagnosis, those who did not engage in follow-up were not included. It is also not clear how adherence was tracked for participants. Two other studies which looked into the impact of marital status on engagement and adherence did not find a link (32, 33).

Patients with a carer were significantly more likely to engage in an initial psychiatry appointment, which acted as a gateway to receiving psychological therapy as part of the CODES trial (COgnitive behavioural therapy for adults with Dissociative non-Epileptic Seizures; 33). As this looked at attendance to an initial psychiatry appointment, the generalisability to adherence to psychological therapy must be questioned. No other studies looked at the effect of a carer.

Putting these quantitative findings into context, qualitative interviews with six people who had engaged in therapy, defined as attending at least one outpatient therapy session, discussed two ways in which their support network had aided their engagement (30). Firstly, family members often encouraged patients to engage. This was especially influential if the family member was a healthcare professional. Secondly, family members often accompanied patients to their initial appointment. This study offers an in-depth understanding of the perspectives of patients with NEAD. However, the study excluded participants who did not engage in psychological therapy, or who did engage but would have likely become distressed by taking part in the study (determined by the healthcare professional). It is therefore likely that this captures the opinions of a smaller sub-group of people with NEAD.

3.3.1.2 Economic factors.

Economic factors were significantly associated with engagement and adherence, as well as being a reason why people do not engage with a psychological intervention.

One study found that being unemployed and on disability benefits was significantly associated with non-adherence (17). Here, adherence was defined as attending at least 8 sessions over 16 weeks, starting at the time of referral. Four other studies looked at the impact of employment status alone and found no significant difference in adherence (14, 17, 32, 33). The Tolchin et al. (17) study also found that lower occupational status was significantly associated with non-adherence. Whilst being unemployed and on benefits, and lower socio-economic status were associated with non-adherence, they did not have predictive power (17). Another study found that having a higher median income (determined by post code) was significantly associated with attending more therapy sessions (36). ‘Financial reasons’ was the second highest rated reason for non-engagement in a study which retrospectively asked participants why they did not engage in psychological therapy (31). Unfortunately, participants were not able to expand on this as data was collected using a non-validated, tick-box questionnaire. It is worth noting that all the studies which linked economic factors to non-engagement and non-adherence were based in the USA, where healthcare is privately funded. Studies which explored the effect of economic factors in the UK did not find employment, disability benefits, or deprivation score to be associated with non-adherence (32, 33).

3.3.2 Healthcare System Related Factors

3.3.2.1 Present Therapeutic Relationship.

Difficulties within the therapeutic relationship were cited as a reason for not adhering to therapy in three studies (15, 28, 29). One of the included studies is a case report of a woman who undertook three sessions of CBT, before dropping out (28). One of the main reasons she discontinued with therapy was feeling as though her therapist was not experienced enough at working with NEAD, leaving her *"at a loss of what to do"* (28, p. 3). She did not feel validated by the therapist for how terrifying it was to experience an attack. As this was a case study of one participant, one must be cautious before generalising these findings. Additionally, there was no analysis on the qualitative data and the paper merely states what the participant said. Similar difficulties were cited in a larger qualitative study, interviewing 30 participants who had taken part in the CODES trial (29). Two participants reported they dropped out earlier on in treatment because they did not feel they had formed an alliance with their therapist. However, this study interviewed participants who had received therapy as part of a research trial. The authors highlight that it is likely that they were more informed about NEAD and the treatment before commencing therapy. Additionally, their motivation to adhere to therapy may have been different to the general population with NEAD, for example engaging in this research trial with a view to helping others with NEAD.

A quantitative study explored participants' views of therapy, and whether they differed depending on if they were adherent to therapy (15). Participants in the non-consensual withdrawal group (defined as deciding to discontinue the program without prior agreement of the mental health caregiver) found the therapeutic relationship significantly less satisfactory than those in ongoing psychological follow-up group, or those who had withdrawn in agreement with their healthcare provider. Additionally, one of the highest cited reasons for dropping out in the non-consensual group of Fettig et al.'s (15) study was *"Lack of confidence in mental health professionals/structures"*. It was not clear why they were dissatisfied as, again, data was collected using a non-validated, tick-box questionnaire.

Positive feelings towards the healthcare system were something that participants reported enhanced engagement and adherence. Several participants in Wyatt et al.'s (30) qualitative interview study commented on the positive ethos of the neuropsychiatry team and how this was valuable in helping participants to engage with therapy. There was no further information on this perceived positive ethos or advice as to how other services can re-create this.

3.3.2.2 Previous experiences with the healthcare system.

Wyatt et al. (30) also found that participants' previous experiences of the healthcare system impacted people's willingness to engage in therapy. Participants spoke about being disbelieved by healthcare professionals they had seen previously, particularly GP's, and the detrimental impact this had on their willingness to engage in therapy. This study had a small sample size of 6 participants, and there is limited information on the interview schedule.

3.3.3 Condition Related Factors

3.3.3.1 Mixed Messages.

Difficulties with engagement and adherence were associated with conflicting messages about their seizures from healthcare professionals in two studies (14, 30). Firstly, participants found it understandably confusing if they were referred for psychological therapy amongst ongoing neurological investigations (30). They were receiving one message that their seizures were neurological, and another message that their seizures were non-epileptic. This uncertainty made engaging with therapy confusing and therefore less likely.

A quantitative study also suggested that patients struggle to engage when they are receiving mixed messages (14). Patients without comorbid epilepsy who were continuing to take anti-epileptic medications were significantly less likely to adhere to therapy. Being prescribed anti-epileptic medication suggests the seizures have a neurological basis, while being referred for psychological therapy implies the seizures have a psychological basis. However, this study was rated as low quality for several reasons, including the fact that inclusion and exclusion criteria for participants was not reported, as well as it not being clear how data regarding adherence was accessed.

3.3.3.2 Effect on Seizures.

A total of three studies found that engagement and adherence is impacted by the effect that treatment has on an individual's seizures (15, 29, 31). One regularly cited reason for not engaging with or adhering to psychological therapy was that patients felt better, or had stopped having seizures (15, 31). Conversely, another study found that an initial increase in seizures could be enough to derail therapy (29). Participants discussed the initial worsening of their seizures due to the higher levels of anxiety caused by beginning therapy, and how this discouraged them from attending future appointments. As previously stated, this was a research trial so generalising this to clinical practice must be done with caution.

3.3.4 Therapy Related Factors

3.3.4.1 Practical Barriers.

One paper found a number of logistical barriers prevented people from engaging in therapy (32). An online CBT group intervention was run by the National Hospital for Neurology and Neurosurgery, London. Participants who disengaged were asked to complete a survey where they were asked to identify the reason for the non-adherence, from a list of options. The majority (14/18) of participants said that they would have liked to have attended but were not able to. They cited reasons such as the groups being too regular and not liking the group format. Some did not like the idea of attending in an online format. This was the only study which explored adherence to a group therapy. A practical barrier cited as a reason for non-engagement in another study was that therapy was too big of a travel commitment (29).

3.3.4.2 Interventions to Address Therapy Related Factors.

A quantitative study addressed issues of therapy related factors by comparing adherence to therapy using telepsychology compared to in-person appointments (36). The telepsychology group had therapy sessions over a video platform. They had their first session in person as this was required by the clinic. Participants chose whether they would like to attend in-person sessions or telepsychology sessions. Comparative analysis of baseline characteristics showed that those in the telepsychology group lived significantly further away. Telepsychology appointments were significantly more likely to be attended and were significantly less likely to be cancelled, even when controlling for potential confounders, including distance lived from the clinic. However, there was one other significant difference between these groups which some of the observed effects may be attributed to. The majority of the in-person therapy was conducted before the Covid-19 pandemic, and most of the telepsychology therapy sessions occurred during the pandemic. The study authors postulate that there could have been pandemic related factors contributing to the increased adherence to the telepsychology sessions. These include having reduced commitments, and hence more time, because of the lockdown, or the fact that people became more health conscious during the pandemic.

Another intervention study explored the impact of the treatment setting on engagement (14). It compared integrated treatment (where psychological therapy and psychiatric management took place in the same clinic) to divided treatment (where psychiatric management took place in the hospital and psychological therapy was undertaken elsewhere). The allocation to different groups depended on where the participant lived. A higher percentage adhered in the integrated treatment group, but this difference was not significant. In this study, different parameters were used to define adherence in the integrated versus divided treatment group. This, along with the potential confounding variable of where the participant lived, makes interpreting the impact of the intervention difficult.

3.3.5 Patient related factors

3.3.5.1 Characteristics.

Some patient demographics appear to be associated with non-engagement and non-adherence. In three studies, the age of participants was found to be significantly higher in the groups which adhered to therapy, both when using the mean (17, 36) and the median (32). However, two studies found no significant impact of age (14, 33). One study looked at cognitive complaints and found that self-reporting cognitive difficulties predicted non-adherence (14).

Three studies investigated whether a history of abuse was associated with non-adherence. Two studies found no link between a history of abuse and adherence (14, 32). In these studies, whether an individual had a 'history of abuse' was determined during the initial neuropsychiatric assessment (14), and from patient notes (32). It is not clear how a 'history of abuse' was defined. Another study found that experiencing child abuse was predictive of non-adherence, defined as attending at least 8 therapy sessions in 16 weeks (17).

Finally, three studies found no impact of ethnicity on engagement and adherence, when comparing if people of different ethnicities were more/less likely to adhere to therapy (32, 33, 36). However, one high quality study found that, when participants defined themselves as being part of a 'racial or ethnic minority', they were significantly less likely to adhere to therapy (17). This self-identified minority status was predictive of non-adherence. The definition of 'self-reported minority status' in the paper was clarified with the author.

3.3.5.2 Disagree with the recommendations.

Two studies reported that participants did not engage because they did not agree with the recommendations, firmly believing that their seizures were epileptic (28, 31). Other studies showed that participants had a general lack of interest in attending therapy (15). In both studies, data was collected from pre-determined options so cannot be expanded upon.

Three studies reported that participants did not wish to undertake CBT specifically (28, 29, 31). For example, participants reported just wanting to have someone to talk to, but had no desire to practice the CBT strategies (29, 31). In one case study, the participant also reported not receiving a rationale of why CBT would be helpful in treating their NEAD, leaving her hesitant to engage (28).

3.3.5.3 Not Feeling Ready.

Participants appreciated that engaging in a psychological intervention could be emotionally challenging and this was something they sometimes did not feel ready for (29,30). They saw therapy as *"simultaneously powerful but hazardous nature"* which caused a reluctance to engage (30, p. 802). Some thought that therapy would be too distressing and did not feel ready to discuss previous traumas (29). This high-quality study involved a thorough Thematic Framework Analysis, allowing nuances within the data such as this to be identified.

3.3.5.4 Interventions to Address Patient Related Factors

To help participants who did not feel ready or disagreed with recommendations, two different types of interventions have been trialled. The first is a Brief Educational Intervention which is based on a previous protocol for communicating the diagnosis of NEAD (38). It was delivered by a nurse and covers topics such as how non-epileptic attacks are psychological in nature and encourages the acceptance of psychological support. This was first formally trialled with 50 participants during an inpatient assessment (35). All of those who had received the intervention had attended or booked a psychological follow-up appointment. Unfortunately, as there is no control group, it is impossible to ascertain the impact of the intervention. Additionally, there is no information given on participant characteristics, or details of how adherence data was obtained. The intervention was later trialled with more sound methodology (34). Participants were randomly assigned to either the intervention or control group, where they were informed of the diagnosis via the neurologist and mental health follow-up was suggested. Significantly more attended or made a therapy appointment in the intervention group. However, there are limitations of this study. The control group does not appear to have received consistent communication of the next steps, and the patients *"may or may not"* have been provided with a follow up referral (34, p. 80). Additionally, it is not clear if the intervention was reviewed for quality and consistency of delivery.

The second intervention is a Motivational Interviewing (MI) intervention, which was assessed with a randomised controlled trial (37). Patients signed up to a 12-session mindfulness intervention. Half of these received an MI session prior to undertaking the mindfulness course and half proceeded straight to the mindfulness sessions. The MI followed a protocol of four steps: engaging, focusing, evoking, and planning. In this high-quality study, sessions were recorded, and the integrity of the MI technique was assessed by two blinded independent reviewers. Significantly more MI participants were adherent, compared to therapy-only participants. Participants who attended at least 8 out of the 12 mindfulness sessions were considered adherent.

3.4 Robustness of Data Synthesis

Sensitivity analysis was performed by excluding the low-quality studies, which were three quantitative studies. The distribution of factors within the WHOMAM's dimensions was re-examined, and there were minimal changes to the results and subsequent conclusions.

4. Discussion

This systematic review collates the current literature regarding engagement and adherence to psychological therapy as a treatment for adults with NEAD. The review identified 12 themes which fit amongst the five dimensions posed by the WHOMAM.

One of the more common reasons cited for not engaging with or adhering to psychological therapy was that patient's seizures had stopped after receiving the NEAD diagnosis. This is common. Around a third of patients report a significant improvement in their NEAD 3-6 months after the diagnosis, with no further intervention (39). It is not seen as a problem that these patients do not adhere to psychological interventions.

While the factors which impact engagement and adherence to therapy spread across the WHOMAM, the evidence for some is more conclusive than others. Having social support and the financial means to undertake therapy (in studies in the USA) enhances engagement. Engagement is more likely if the communication of the diagnosis is clear, an effect observed across Functional Neurological Disorders as a whole (40). Many patients simply do not feel ready to engage in psychological therapy or saw it as an investment that comes with associated risk. One of the main reasons cited for not adhering to therapy was a poor therapeutic relationship. It appears that patients engage more when therapy is conducted on the same site as the patient's psychiatry appointments or having the option of online sessions. Motivational interviewing and educational interventions were shown to significantly increase adherence to therapy. As with studies into adherence for psychological interventions more generally (41), older patients seem more likely to adhere to psychological therapy.

4.1 Limitations of Included Studies

A re-occurring limitation was that data was collected with non-validated and closed questionnaires, which asked participants to tick a reason as to why they did not adhere to therapy. Whilst this made participating less demanding and therefore may have increased recruitment, it limits the information that can be derived from these studies.

There was also significant variation between included studies, which makes directly comparing studies difficult. The way in which included studies defined engagement and adherence differed significantly, although this is common in systematic reviews of adherence (41). The studies examined adherence to a number of different psychological interventions. Some studies did not include information on the psychological approach taken.

This review is heavily dominated by research from higher income countries. Research has found a number of differences in how treating NEAD is approached in high-income countries compared to low-income countries (42). Therefore, it may be that the factors associated with engagement or adherence found here do not generalise to lower-income countries. Additionally, the differences in how healthcare is funded across different countries was likely reflected in some of the barriers found to engaging in therapy.

Finally, both randomised control intervention studies had a control group of treatment as usual. In both, more time was spent with the patients in the intervention group (30-90 minutes). In a population that feel healthcare professionals do not listen to them, it is possible that it was this extra time spent with them which increased adherence, rather than the intervention itself.

4.2 Strengths and Limitations of this Review

This review provides a comprehensive overview of the current literature regarding engagement and adherence to psychological therapy as a treatment for adults with NEAD. It employs systematic methodology and uses an evidence-based model, the WHOMAM, as a framework for conceptualising the findings. The inclusion of quantitative and qualitative studies provides evidence into what factors are associated with engagement and adherence, as well as contextual information as to why this is. It is important for systematic reviews to include grey literature, to try and minimise publication bias (43). This review includes an unpublished thesis (31). Additionally, the authors of conference presentations were contacted and asked for further information to enable us to include these, a valid Cochrane methodology (44). Unfortunately, none of the authors responded.

Nevertheless, the review has some limitations which have been considered within the recommendations outlined by Busse et al. (27). There are several different terms for NEAD, including psychogenic, functional, and dissociative seizures (45). The primary author did their best to include all possible terms in the search strategy, but it is possible that some were missed. Studies were excluded if they explored factors which could be linked with engagement or adherence but did not make this link explicitly. For example, one qualitative study explored the *"blame and humiliation"* that patients felt from their previous healthcare providers and the authors hypothesised that this would impact willingness to engage in future treatment, but this was not asked of participants (46, p. 10). Additionally, some opinion pieces spoke to the topic, for example the role of shame (47), but these were not included in this review as they were not empirically grounded. Finally, while using the WHOMAM to guide the synthesis has its advantages, the distinct categories provided by the model do not allow for overlap between the factors to be explored.

4.3 Conclusions and Future Research

The factors associated with engagement and adherence to psychological therapy for adults with NEAD are multi-factorial and span across the WHOMAM. While there is more research that explores patient-related factors, this does not mean these factors are the most important. It is likely that the most effective interventions will target several dimensions of the model, for example giving a clear diagnosis, providing an MI intervention targeted at younger patients, while also offering telepsychology appointments. An individual's support system can also be leaned upon to increase engagement and adherence. Further research into multifaceted interventions would be valuable. Additionally, future intervention studies should control for the time given to the patient by a healthcare professional, so that a more confident conclusion can be drawn about whether the intervention itself increases engagement and adherence. Some factors which have been associated with engagement and adherence to psychological therapy in other populations, such as duration of the problem and who referred an individual for therapy (41), were not considered by any of these studies, so may be an area for future research. Eleven out of the 13 studies included in this review were published in the last 10 years which shows what a developing and exciting area of research this is.

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Service Improvement Project

Investigating the decision-making process regarding surgery for parents of children with craniosynostosis at the Oxford Craniofacial Unit.

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Proposed Journal

The Cleft Palate Craniofacial Journal is an interdisciplinary, international journal dedicated to research in all areas of craniofacial conditions. It has recently published qualitative research exploring the psychological impact of craniofacial conditions on individuals. Author submission guidelines are presented in Appendix E.

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Declaration of Interest

There are no conflicts of interest.

Abstract

Objective: This Service Improvement Project explored the current decision-making process regarding corrective surgery for Craniosynostosis; a decision made by parents/carers on behalf of their children. The study examines what information parents use to make this decision, as well as exploring what is being done well, and what could be improved within the service.

Design: Online qualitative surveys were used to collect data.

Setting: The study was conducted at one of the five highly specialised Craniofacial centres in the UK.

Participants: Eighteen parents of children with Craniosynostosis and ten MDT members participated in the project.

Main Outcome Measure(s): Qualitative surveys were analysed using Thematic Analysis. The parent and MDT surveys were analysed separately. Analysis was guided by the Theory of Reasoned Action and Theory of Planned Behaviour.

Results: The analysis showed that the MDT are promoting shared decision making across the team. Parents base their decision on a number of factors, including their risk tolerance, values around appearance, and the opinions of others (e.g. society and family/friends). The parents discussed the challenges of the decision and the associated difficult emotions, but praised the support offered by the Craniofacial Team. Suggested improvements to the process are detailed.

Conclusions: The decision about corrective surgery is an emotive experience for parents, however this is a process parents feel supported with by the Oxford Craniofacial Team. Suggestions to improve this process for parents even further are listed.

Keywords: Craniosynostosis, shared decision-making, service improvement

Introduction

Craniosynostosis

Craniosynostosis is characterised by the premature fusion of one or more cranial sutures in a baby's skull (Garza & Khosla, 2012). This often results in an abnormal skull shape (Speltz et al., 2004). It is a rare condition, which affects between 1 in 2,000 and 1 in 5,000 infants a year (Kolar, 2011). Ninety-five percent of cases are non-syndromic, meaning the cause is unclear. Risk factors include prenatal exposures to certain medications and too little amniotic fluid during pregnancy (Johnson & Wilkie, 2011). If the skull shape is not corrected, there is a 15% chance of developing raised intracranial pressure in children with non-syndromic craniosynostosis. If this is not promptly addressed, it can lead to brain injury and delayed development (Eide et al., 2002). There can also be social and emotional consequences of craniosynostosis as children may be bullied due to their visible difference (Ozgur et al., 2006).

Corrective Surgery

Surgery to correct the infant's skull shape is an important part of treatment (Kluba et al., 2016). However, this major surgery comes with risks of complications, including intraoperative haemorrhage and significant blood loss (Boop et al., 1996). The chances of these complications occurring depends on a number of factors and are therefore different for each child (Lee et al., 2012). These risks increase with age, and surgery usually takes place before the child is 18-months (Bruce et al., 2018). To minimise these risks, surgery is only performed at five highly specialised centres in the UK (Care et al., 2019). Children with non-syndromic craniosynostosis generally perform in line with their peers across developmental, behavioural, and emotional domains (Care et al., 2019, 2021). However, this sample was drawn exclusively from children who have undergone corrective surgery. Systematic reviews paint more of a mixed picture of developmental outcomes, and the quality of the evidence is mixed (Knight et al., 2014).

Decision-making

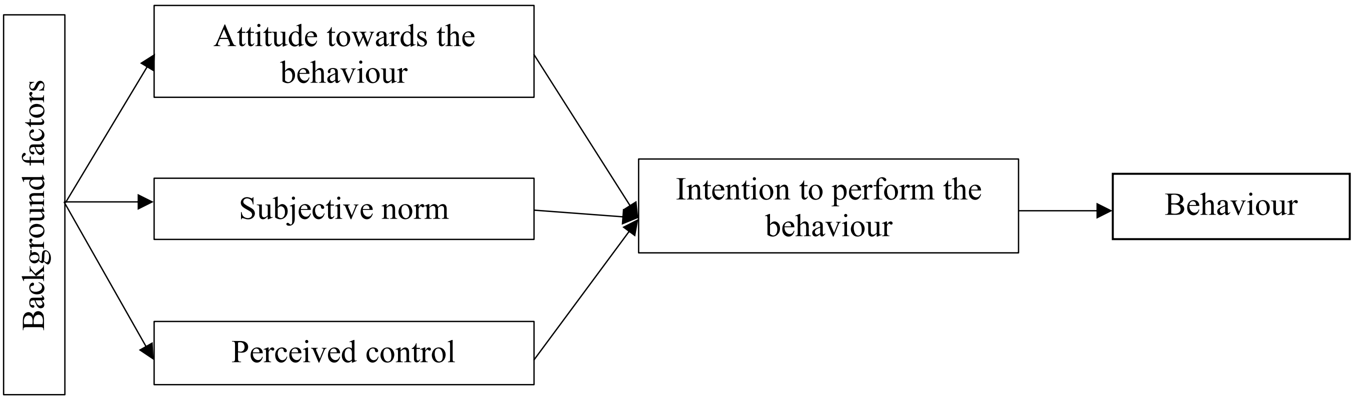
Parents must decide on behalf of their child whether to proceed with surgery, considering both functional and cosmetic elements in the absence of thorough longitudinal research. At the Oxford Craniofacial Unit, where this study is based, around 90% of parents choose for their child to undergo corrective surgery. There is limited research into what parents base their decision on. A Swedish study found 30.8% elected for surgery based on aesthetics, 38.5% feared brain damage, and 23.1% cited a combination (n=26) (Gewalli et al., 2001). Despite advice from health authorities, there have been no UK-based studies into understanding and improving decision-making support in Craniosynostosis (Rasmussen et al., 2008).

In the broader literature on medical decision-making, the integrated theories ‘Theory of Reasoned Action’ and ‘Theory of Planned Behaviour’ (Ajzen, 1991) are commonly used to explain decision-making processes. The combined theories explore how an individual’s attitude, opinions of those around them, and perceived control over carrying out the behaviour influence the decision to engage in that behaviour (Figure 2.1). In the current study, the ‘behaviour’ in question is the decision to consent to surgery.

‘Attitude towards the behaviour’ is comprised of instrumental attitude (cognitive beliefs about the behaviour or outcome) and experiential attitude (emotions towards performing the behaviour). ‘Subjective norm’ consists of the beliefs of other important individuals and an individual’s motivation to comply with these. ‘Perceived control’ refers to an individual’s sense of self-efficacy and whether they possess the abilities needed to perform the behaviour. The model recognises several external variables that can influence each of the three aforementioned domains. It has been used to explore medical decision making in other conditions, such as breast cancer (Sivell et al., 2013).

Figure 2.1

Theory of Planned Behaviour and Theory of Reasoned Action (Azjen, 1991)



The concept of shared decision-making is becoming increasingly important in the NHS since Andrew Lansley coined the expression “*no decision about me, without me*” (Lansley, 2011). Shared decision-making involves providing patients with evidence-based information about potential benefits or harms of an intervention (Coulter et al., 2011) and has been shown to lead to improved child health status across paediatric settings (Park & Cho, 2018). Shared decision-making is recognised in the NHS Long Term Plan as a way of improving patient-centred care (NHS, 2019).

Questions to be Addressed

Considering the above, the Craniofacial service at Oxford is seeking to understand the current decision-making process for parents within their service, to ensure the right support is in place to help make this process as smooth as possible. This Service Improvement

Project is guided by the Plan, Do, Study, Act (PDSA) cycle suggested by NHS England (NHS England, 2021).

The following questions will be considered:

- What are parents' experiences of decision-making regarding surgery?
- What made the decision easy/difficult?
- What is currently working well in the decision-making process?
- Could specific changes be made to the service to improve this experience?
- How do members of the MDT currently ensure there is shared decision-making?

Methods

Setting

The study was based in the Oxford Craniofacial Unit, one of five highly specialised craniofacial units in the UK. It took place over an 8-month period, starting in April 2022. The service cares for children with Craniosynostosis across the south of England. For 2021-2022, the service received 199 referrals. During this year, the service performed 77 operations.

Participants

MDT

All health care professionals working in the Craniofacial service during the study period (N=18) were invited to complete a survey. The MDT includes surgeons, a clinical nurse specialist, clinical psychologists, speech and language therapists, orthoptists, and clinical geneticists. A Psychologist in the team was excluded as they supervised the project. The survey was sent out via email, discussed in the team meeting, and reminders sent. The decision was made to not ask participants to disclose their profession, as this could have made them identifiable.

Parents

Parents were approached at their child's routine 3-month post-surgery appointment. If the family consented, an email was sent with a link to the survey. The decision was made to only have one parent from each family unit participate, so the results were not skewed by some family units having more participants than others.

Survey Development

Two surveys were created, one for the MDT and one for parents. Draft survey questions were based around the Theory of Reasoned Action and Theory of Planned Behaviour, and relevant literature.

Feedback on the draft MDT survey was obtained from a Clinical Psychologist who had previously worked in the Craniofacial Team. The wording of questions was altered in line with their feedback. The final survey consisted of 12 questions (Appendix F).

Feedback on the draft parent survey was sought from the NIHR Cleft and Craniofacial Clinical Studies Group, which includes people with lived experience of Craniosynostosis. Recommendations put forward by the group were incorporated. The final survey consisted of 13 questions (Appendix G).

Ethical Approval

The study was deemed 'Service Evaluation' by the Oxford University Hospitals (OUH) CTRG and therefore ethical approval was not needed. In line with hospital guidelines, the project was logged with OUH.

Data Analysis

The MDT and parent data were analysed separately, using Reflexive Thematic Analysis (Braun & Clarke, 2006, 2013), conducted using NVivo. As per the Thematic Analysis guidelines, the primary researcher began by familiarising themselves with the data. Next, the primary researcher systematically coded each survey response. Coding was done using a semantic, inductive approach, as the codes were generated solely from the data. At the end of the coding process, the MDT data had 60 codes and the parent data had 53 codes.

Next, the researcher constructed themes from the coded data, guided by the Theory of Reasoned Action, and Theory of Planned Behaviour. As this was guided by theory, this stage of the analysis used more of a deductive, top-down approach. The researcher mapped the codes onto the theory where possible. If there were patterns within the data which were not accounted for within the model, a new theme was created. This enabled the researcher to be guided by an evidence-based model, while allowing some flexibility. Themes were revised, named, and described once the researcher was satisfied that they captured the patterns evident in the data.

On the ontological continuum, the researcher takes a critical realist approach, acknowledging that knowledge is socially influenced and, as researchers, we can only ever access this level of socially-located knowledge (Braun & Clarke, 2013). Epistemologically, the researcher takes a contextualist stance. Within contextualism, the researcher retains an interest in understanding the ‘truth’ but sees knowledge as impacted by context and the researcher’s position, biases, and assumptions (Madill et al., 2000).

Given this, a codebook was not used as the researcher is not aiming for objectivity. Subjectivity was seen as “*a resource which can be tapped in order to contextualise and enrich the psychological research process*” (Gough & Madill, 2012, p. 1). The primary researcher practiced reflexivity by conducting bracketing interviews (Appendix H), to understand any biases or assumptions throughout data analysis (Rolls & Relf, 2006). Areas explored in this interview included the researcher imagining herself as a participant and discussing how she may answer the questions (Gough & Madill, 2012). Additionally, a reflexive journal was kept throughout the study.

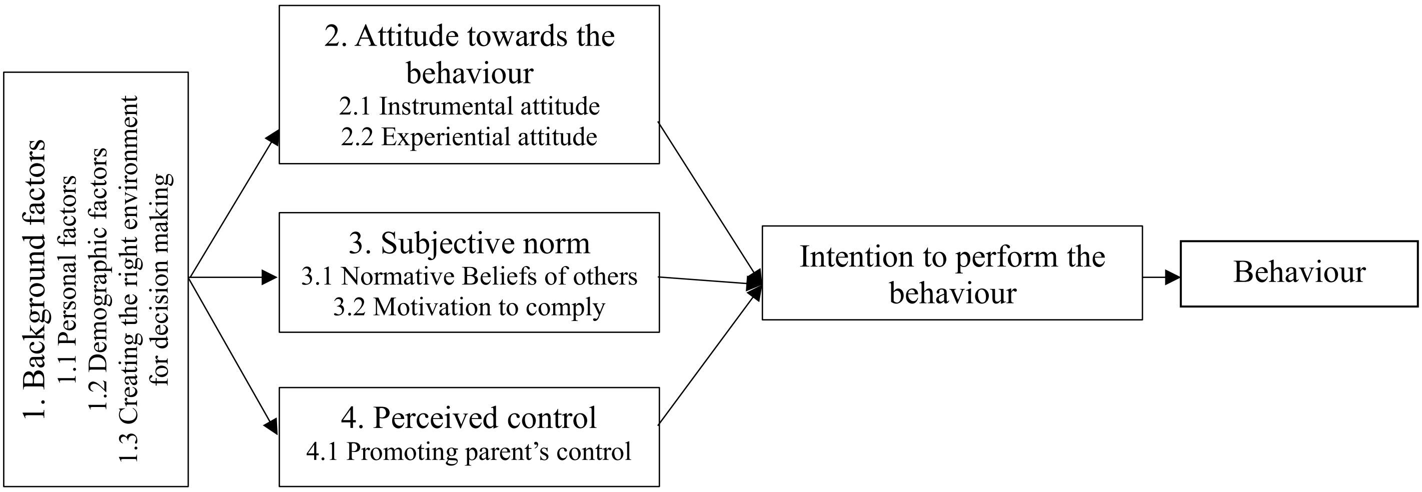
Results

MDT Analysis

Ten participants from the MDT completed the survey. In the MDT, data 4 themes and 9 sub-themes were identified. The themes and sub-themes are numbered in Figure 2.2, and expanded upon below. A summary of themes, sub-themes and codes can be found in Appendix I. Additional quotes from each theme and sub-theme can be found in Appendix J.

Figure 2.2

MDT Themes



1. Background Factors

The MDT participants discussed how several background factors impacted parents' decision making. These mean that each family's consultation is tailored to them, and they endeavour to *"keep the individual child at the heart of the consultation"* (MDT1). These individual differences are expanded upon below.

1.1 Personal Factors

MDT members felt parents and families differed on their attitudes towards risk. MDT5 stated *"what is an acceptable level of risk for one set of parents, which [sic] may totally differ to another"*.

Respondents thought that parents also differed on their values around appearance. These differing values were often shaped by the parents' past and any previous positive or negative experiences regarding appearance. MDT10 stated *"if a parent has their own experience or is aware of other's experiences of negative reactions around craniofacial difference, they may feel that they want to prevent this from happening to their child by opting for surgery"*.

1.2 Demographic Factors

MDT members appreciated the impact of demographic factors, and how intersectionality of different characteristics could impact parents' decisions.

> *"I do think there is some intersectionality with race/ethnicity in terms of making the decision. For example, one father was particularly worried about his son, who was black, growing up with a scar on his head and how this may affect his future job prospects."* (MDT3)

1.3 Creating the Right Environment for Decision Making

The MDT members discussed how clinic environments can make taking on board information difficult. They discussed how parents' initial discussion with the MDT is in clinic *"which is quite a stressful environment for parents, with lots of information given, and lots of professionals in the room"* (MDT9). Additionally, *"the time constraints of NHS appointments are difficult"* (MDT1), and the team are working within *"a window for a transcranial procedure (e.g. before children's sinuses develop)"* (MDT 1).

Participants spoke of efforts to create an environment more conducive to decision making. Several participants discussed how having multiple appointments with the MDT can help parents to take in the information they need. They discussed the importance of validating and normalising difficult emotions, and often this is where Clinical Psychology come in; *"I do think the pre-operative psychological input can really make a difference to the parents experiences, even if they don't know what to expect going in"* (MDT3). As well as input from individuals, the whole MDT aim to be *"offering a reflective, non-judgemental, safe space to explore options and sound out thoughts and ideas"* (MDT5) throughout the decision-making process, where it is *"ok for parents to accept or decline offered surgery"* (MDT5).

Some reflected that creating a helpful space for parents to think through their decision is something that the team could improve on. For example, MDT9 suggested; "*I wonder whether there is any way parents could have an opportunity to discuss surgery and their questions without all of us there?"*.

2. Attitude

Attitude can be broken down into an individual's instrumental attitude and experiential attitude about performing a behaviour. Within the MDT data, there was significantly more discussion regarding elements of parents' instrumental attitude, indicating that the team perhaps see factual information as more central to decision making.

2.1 Instrumental Attitude

The MDT participants reported that parents consider a large amount of information during the decision-making process. Parents often envisaged what their child's future might look like, proceeding with surgery or not. The risk of raised intracranial pressure was often thought to be the deciding factor; *"I think that once the risk of high pressure is mentioned, this makes the decision 'easier' for the parents"* (MDT2).

MDT members aimed to communicate clear, detailed, and evidence-based information in a consistent manner. They felt it was important to have *"all members singing from [sic] same hymn sheet"* (MDT6). They also thought it was important to discuss options provided by other specialist centres and what follow-up care will look like whether they proceed with surgery or not.

The consensus from the MDT was that *"pre and post op photos [sic] very useful for placing into perspective"* (MDT4). Additionally, they thought that visual or written information could be useful; *"A written summary of information could help provide a balanced view that families can refer to at a later date and share with others to help them make the decision"* (MDT3).

Respondents felt that parents often sought additional information from other sources such as peer support from fellow parents via social media, information from charities such as Headlines, or via Google. The latter caused concern amongst the MDT; *"I'm sure most people will search Google images which can produce some extreme cases, and some inaccurate information"* (MDT3).

Bringing the above information together and ensuring that parents have understood the pros and cons for their child often fell to the specialist nurse. For example, MDT9 stated *"[specialist nurse] is really approachable and available to answer lots of questions following clinic appointments"*. The MDT as a whole saw their role as supporting parents to weigh up the pros and cons of surgery and approach the decision-making process in as logical a way as possible.

2.2 Experiential Attitude

MDT members reported they *"often see how this decision weighs on parents' minds"* (MDT3). Additionally, *"parents feel the pressure to make the right decision for their future, which is a big responsibility"* (MDT3).

MDT members felt that parents found the decision harder in non-syndromic cases, where there is very little visible difference and a lower risk of raised intracranial pressure. This was made harder when the MDT do not recommend a particular decision.

3. Subjective Norm

3.1 Normative Beliefs of Others

Respondents reported that parents often talked the decision through with each other, and with their closest family members. One MDT participant therefore questioned whether the decision is harder for single parents who may be more isolated.

Participants discussed how many parents often wish to seek their child's opinion, something which is not possible. MDT1 explained that *"the agonising part of surgical decision making for parents is trying to make a decision that they would like to have their child's voice and views on, when their child is too young to participate"*.

It was also reported that parents often sought out the opinion of the MDT, but a strength of the MDT is to promote shared decision-making, and *"even though some parents would like us to make the decision for them, we resist, and encourage them to take some ownership of it, with our support"* (MDT6).

3.2 Motivation to Comply

The MDT members spoke about the value which society places on appearance, which influences parents' decisions. Their motivation to comply with these societally imposed standards differs between parents but is something which can often feel uncomfortable.

> *"The primary function of the surgery is to allow their child to conform with societal norms which can be an uncomfortable position to take as a parent. I think thoughts of "why should I force my child to go through a serious surgical procedure just so they'll fit in?" are often contrasted with thoughts of "who am I to deny my child their right to a 'normal' childhood?""*
> (MDT3)

4. Perceived Control

4.1 Promoting Parent's Control

Developing trust between parents and the MDT was key to helping parents feel in control of the decision; *"establishing a strong rapport and clinical relationship with the patient's family is key"* (MDT8).

Getting alongside parents and understanding their values and beliefs enabled them to help parents' voices to he heard by ward staff or the wider MDT. MDT10 stated *"I may also advocate for a family, representing their views to the wider MDT"*.

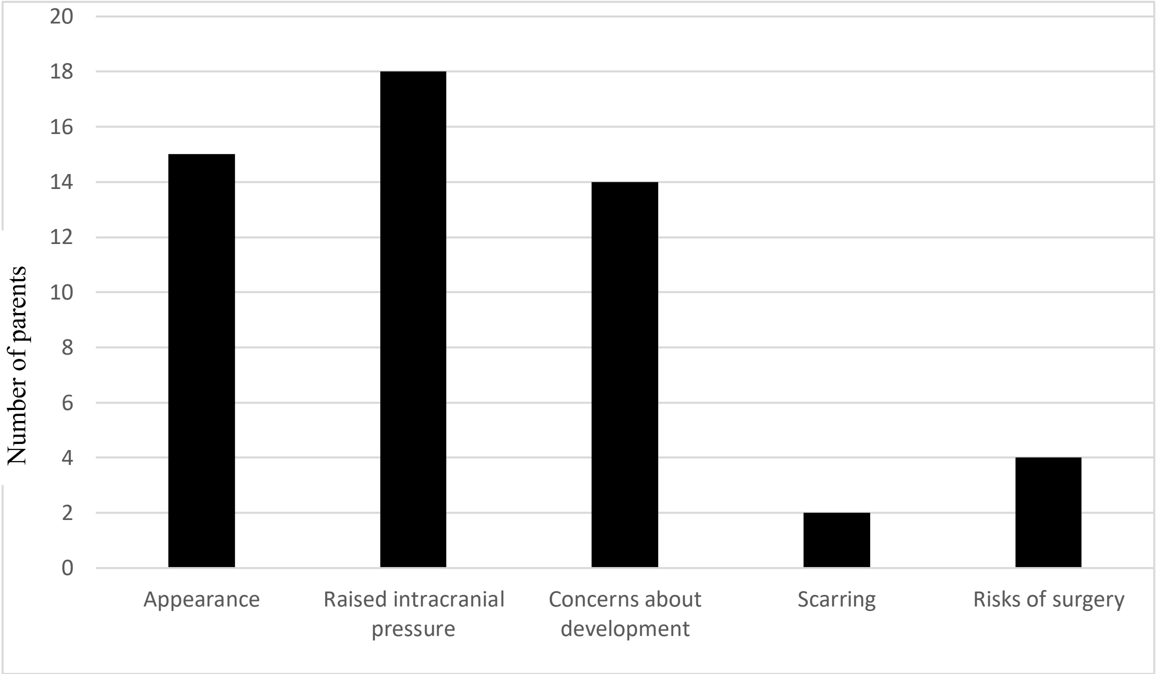
Parent Analysis

Thirty-eight parents were approached and agreed to participate in the study. A total of 18 parents completed the survey. As surveys were emailed to parents and completed anonymously, data is not available to indicate reasons why parents did not participate. Sixteen parents were female, two were male. Twelve had sons and six had daughters. The median age of their child currently was 23.5 months. The median age of their child's first surgery was 17.5 months. Some of the children had other diagnoses, including hernias, developmental delay, and eczema.

Parents were asked which factors they had considered when making the decision about whether to pursue surgery. The results of this can be seen in Figure 2.3. Raised intracranial pressure was noted by every parent as a factor.

Figure 2.3

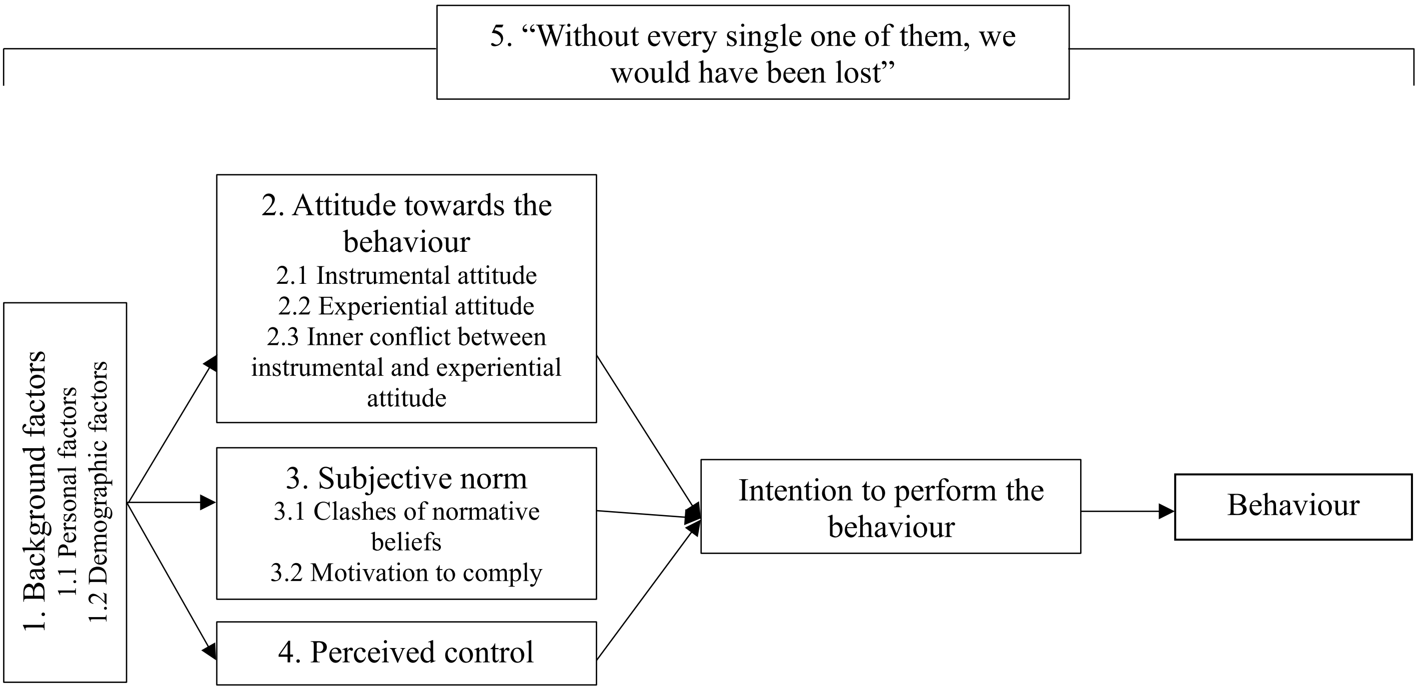
Responses to parent Question 2 (What guided your thinking about surgery as an option? Please tick all of the following options which apply.)



In the parent data, 4 themes and 7 sub-themes were identified within the Theory of Reasoned Action and Theory of Planned Behaviour, and one theme was identified outside of this. The themes and sub-themes are numbered in Figure 2.4, and expanded upon below. A summary of codes, sub-themes, and themes can be found in Appendix K. Additional quotes can be found in Appendix L.

Figure 2.4

Parent Themes



1. Background Factors

Parents thought that the MDT effectively considered their differing background factors and life circumstances; *"Not once have I ever felt my daughter was 'just another patient'"* (P1).

1.1 Personal Factors

Parents differed in their willingness to tolerate risk, for example P3 stated *"our son might never experience issues caused by his head shape, but we considered this to be a* *gamble we weren't prepared to make"* (P3). Attributions around previous experiences were also important. For one family, having a negative experience of previous surgery for their child made the decision regarding craniofacial surgery harder. For another family, a positive previous experience made the decision easier.

1.2 Demographic Factors

The child's gender influenced parents' decisions some of the time. For example, P1 stated that she worried more about her child being bullied for her appearance, because she's female.

2. Attitude

2.1 Instrumental Attitude

Parents considered a wealth of different information when making the decision regarding their child's surgery. Raised intracranial pressure was seen by all parents as the most important factor; *"the Oxford team explained about the risk of inter [sic] cranial pressure that was the key point for us in agreeing to go forward with surgery"* (P16).

Parents sought to gain a clear and complete understanding of the option of surgery. Further tests helped to clarify elements for some parents. Most parents found looking at pictures of before and after surgery very useful. P5 discussed how there were *"no pictures of children older than 5 or 6, no pictures of children who chose not to have surgery at an older age & nothing to show teenagers or adults which is what we wanted to see"*. Ultimately, parents discussed wanting to avoid a state of hypervigilance as their child grows up. P5 stated that *"we would be worried every time he had a headache or had an episode of staring if we did not operate"*. Several parents explained how proceeding with surgery felt like the only option.

Parents found this information through a variety of routes. Several parents were very appreciative of the fact that the MDT "*were happy to answer any questions with honesty & discuss any concerns whether it be during appointments or via email*" (P15). Parents often did their own research, whether this was reading Care Quality Commission reports, or watching documentaries. Discussing the decision with other parents of children with Craniosynostosis was particularly helpful for some parents; *"I would strongly recommend providing the opportunity for parents to speak to other parents that have been through the same process"* (P3). Some parents sought this connection through Facebook groups; however, this was not always a positive experience. P16 stated that *"these were quite anxious places that weren't helpful for us"* and some *"found a lot of irrelevant information which made things very confusing"* (P15). In light of this, some thought a decision aid which documented the relevant information would have been of use.

2.2 Experiential Attitude

Parents described a *"huge rollercoaster of ups and downs"* (P8) in their emotions towards making the decision to put their child through surgery. Parents describe feeling *"anxiety, impatience"* (P10), *"pure fear"* (P17), and *"stress and anger"* (P3). One parent discussed how she *"ended up needing professional trauma therapy"* (P1). Surgery going wrong was something that weighed on parent's minds, and they feared losing their child during the surgery. They often cited feeling pressure to make the right decision for their child. One of the most challenging aspects for parents was the thought of *"exposing our completely healthy son to a procedure that could result in paralysis, blindness, disability and death"* (P3).

The difficult emotions experienced were made worse by the possibility of surgery being cancelled, as well as the delays in receiving a diagnosis caused by the Covid-19 pandemic. P16 stated that *"we realised [child] had an issue at a few days old but due to covid and referral delays didn't meet the Oxford team until [child] was around 5 months"*.

2.3 Inner Conflict Between Instrumental and Experiential Attitude

Several parents spoke about the difference between thinking about the surgery from a rational point of view, and from an emotional point of view. Whilst the decision to pursue surgery was somewhat 'easy', the emotions brought by proceeding were difficult to tolerate. This is demonstrated by P3 who stated *"the rational element of the decision-making process was relatively easy, overcoming the emotional barriers was difficult"*.

3. Subjective Norm

3.1 Clashes of Normative Beliefs

Where there were two parents present, parents valued each other's opinions on whether to proceed with surgery. Some parents worked through this decision together, while others reached a decision independently before conferring with the other. When there was only one parent responsible for the decision, P12 described how the decision was *"all on me"*.

Parents often discussed how society's values around appearance often clashed with their own and this subsequently impacted their decision making. Many feared their child being bullied.

> *"As part of our parenting approach we also want our children to understand that physical looks don't define who you .. The wider world (e.g. school peers potentially bullying in future) wouldn't all share this perspective and so that element of the aesthetic issue did feel more important." (P16)*

Finally, family members' thoughts on the decision regarding surgery were often offered, to parents, whether requested or not. Participants spoke about how *"it did feel at times like people were questioning our judgement as parents"* (P16).

3.2 Motivation to Comply

Interestingly, while parents often talked this decision through with their family, parents had minimal motivation to comply with their family's beliefs if they differed from their own because they felt that they knew what was best for their child. For example, P7 stated that *"we spoke to both our families, but it wasn't changing out [sic] decision we just needed to talk it over further with other people"*.

4. Perceived Control

Parents consistently stated that the MDT *"were always very clear we had a choice about surgery"* (P16) and "*our thoughts and feelings were always at the heart and we were never rushed into making a decision"* (P5). In terms of making the final decision, this was done at very different points in the process for different families. Some parents decided to proceed with surgery *"the same day we were advice [sic] that surgery was on the cards"* (P17) and others *"questioned right up to the last hour if it was the right thing to do"* (P15).

5. "Without Every Single One of Them, We Would Have Been Lost" (P10)

This theme relates to the sense of gratefulness which was expressed by a number of the parent participants. In Figure 2.4, the sub-theme has been placed above the theory to reflect the fact that parents were grateful for the MDT's efforts throughout different elements of the decision-making process. They adapted to an individual family's needs, attitudes, previous experiences, and preferences.

Throughout the parent survey responses, there was a clear theme of how grateful parents were to the MDT, mentioning many of them by name. They were described as caring, exceptional, clear experts, and confident. Parents thought that the MDT *"handled our stress very well, were patient, answers [sic] all the questions with all their experience and* *knowledge. We have met right people in the right place. We can't thank them enough!!!"* (P11). Parents discussed how you must *"put you [sic] child's life in someone else's hands"* (P10), but they felt able to do that with this team and *"trust complete strangers"* (P18). Within the MDT data, participants spoke about creating the right environment for decision making, by providing opportunities for reflection and discussion, over multiple appointments if needed. This is something that parents appreciated.

Discussion

Throughout both the MDT and parent data, there was strong evidence of shared decision-making being present throughout the process (Lansley, 2011). Within the paediatric shared-decision making literature, there is very little research into the perceptions of medical professionals (Carlisle et al., 2023). The current study adds to this and shows that the Craniofacial MDT prioritise shared decision-making. They aim to provide evidence-based, clear, and detailed information regarding the pros and cons of surgery, whilst making it clear that the ultimate decision lies with parents. Nurses' role within this in helping parents to understand and weigh up the options has been noted in previous research (Park & Cho, 2018) and was spoken about by both MDT members and parents in the current study.

Some, but not all, MDT and parent participants thought that a decision aid would be helpful. Research has established that when it comes to communicating risk in paediatric settings, visual or written tools are the preference of most parents (Selvarajan et al., 2023).

Whilst MDT members will often support parents to make a decision, parents were clear that the ultimate decision lay with them. For some, this decision was easy and immediate. For others, this decision was painstaking and did not feel final until consent was taken on the day of surgery. Echoing previous literature into parental decision making in paediatric settings, this process is made easier by the presence of trust, collaboration, and mutual respect between parents and healthcare professionals (Park & Cho, 2018). MDT members strove to create an environment with the aforementioned qualities. They did this by being patient in answering parents' questions, seeing parents over multiple appointments if needed and creating a place for discussion, emphasising that there is no right or wrong decision about surgery. Parents were exceptionally grateful for this.

The analysis details the significant number of factors which are taken into consideration when parents make this decision, which are shaped by a parents' background and life experiences. In line with the limited previous research in this area, parents considered the possibility of raised intracranial pressure to be the most important factor to consider (Gewalli et al., 2001). Seeing photos of children before and after surgery helped parents in this process (Care et al., 2018).

Implications for Service Improvement

The following recommendations have been developed to improve the decision-making process in the Oxford Craniofacial Team.

1) To explore a process of putting parents currently considering surgery for their child in touch with consenting parents who have recently been through the decision-making process.
2) To look into whether it is feasible for parents to meet with Nursing and/or Psychology after their initial appointment where surgery is discussed to clarify their understanding of all the relevant information.
3) For all MDT members to be mindful of single parents who may have limited opportunity to talk this decision through with someone close to them.

4) Where possible, to incorporate pictures of older children (who did and didn't opt for surgery) into the pictures shown in clinic to give parents an insight into how their child might look as they grow older.
5) To develop a decision aid document or written summary of the clinic appointment to aid with retention and sharing information with significant others.

Strengths and Limitations

The current study has several strengths. An evidence-based theory (Theory of Reasoned Action and Theory of Planned Behaviour) was used throughout the study, from designing the questions through to interpreting the data. This theory driven approach helped to guide this project which was advantageous given the distinct lack of other research in this area. Additionally, incorporating both parent and MDT perspectives gives a clear insight into the decision-making process from both sides. Finally, experts by experience were consulted in the development of both the parent and MDT survey to ensure that the language that was used was acceptable.

In terms of limitations, one could argue that both participant groups were somewhat biased. All MDT members were invited to participate but it is possible that only the ones interested in this area completed the survey. By recruiting at a child's post-operative assessment, we were only able to recruit those who proceeded with surgery. Caution must therefore be applied when generalising these findings to parents who decided for their child not to have surgery. Additionally, it is possible that parent's survey responses were impacted by recall bias and may have been effected by the fact that their child had come through surgery. Furthermore, the majority of parent responders were mothers, so caution must be taken when applying the findings to fathers or other carers. In terms of the analysis methods, while being guided by theory has its advantages, this meant that themes were not truly data driven. It is possible therefore that this constrained some of the analysis and links that would have otherwise been made within the data if an inductive, bottom-up approach had been used throughout.

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Theory Driven Research Project

Exploring the Impact of Perceived Social Support as a Moderator of the Relationship Between Perceived Public Stigma and Negative Outcomes in Bell's Palsy

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Proposed Journal

Body Image is an international, peer reviewed journal which presents scientific articles on appearance research. It welcomes papers on the impact of physical characteristics on psychological functioning, among other things. Author submission guidelines are found in Appendix M.

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Declaration of Interest

Declarations of interest: none.

Abstract

Objective: The main aims of this study were to explore the differences in perceived social support between people with Bell's Palsy (BP) and the general population, and to test for a potential association between perceived stigma and psychological distress in the BP population. Perceived social support was tested as a potential moderator of this relationship.

Design: This study employed a cross-sectional survey design.

Participants: Ninety-four people with BP and ninety-one controls without BP participated in this study.

Main Outcome Measure(s): Perceived Stigma Questionnaire, Multidimensional Scale of Perceived Social Support, Patient Health Questionnaire-8 (depression measure), and Generalised Anxiety Disorder Assessment-7.

Results: People with BP reported significantly lower levels of perceived social support and higher levels of depression and anxiety compared to people in the general population. Perceived stigma was significantly associated with (and predicted) negative psychological outcomes, but perceived social support did not moderate this relationship.

Conclusions: Limitations of the study are discussed. Future research should explore other potential moderators of the relationship between stigma and psychological outcomes.

Keywords: Bell's Palsy, social support, stigma, anxiety, depression

Highlights:

- Bell's Palsy
- Social Support
- Stigma
- Anxiety
- Depression

Introduction

Facial Palsy

Facial Palsy (FP) affects around 100,000 people in the UK (Facial Palsy UK, 2012). It is characterised by a temporary or permanent weakness in the facial muscles which adversely impacts both appearance and function (Benecke, 2002).

FP can be congenital or acquired. Research has found that those with acquired FP have higher rates of anxiety and depression, compared to those with congenital FP (Bogart, 2020). Bell's Palsy (BP) is the most common type of acquired FP, thought to be caused by a viral infection (Tiemstra & Khatkhate, 2007). Approximately 70-80% of people who experience BP recover in a few months, however for some the facial weakness is permanent (NICE, 2019). Treatment involves receiving injections of botulinum toxin A (Filipo et al., 2012), engaging in physiotherapy (van Landingham et al., 2018), or undergoing reanimation surgery (Ghali et al., 2011).

The changes in function and appearance of the face, along with an uncertain recovery pathway, can result in anxiety and depression (Norris et al., 2019). Some of the literature indicates higher rates of anxiety and depression, compared to controls (e.g. Pouwels et al., 2016; Walker et al., 2012), others found no difference (e.g. D. Sun et al., 2015). To date, psychological interventions have typically focused on self-help approaches incorporating elements from a range of therapeutic models (Hotton et al., 2022).

Impact of Facial Palsy on Social Interactions

Social interactions are affected by FP in two ways. Firstly, a lack of facial expression can be misinterpreted by others. The human face serves a crucial role in social interaction by providing non-verbal cues regarding a person's emotion (Frith, 2009; Horstmann, 2003). Facial expressions communicate the seven universal emotions crossculturally (Ekman et al., 1969). Not only does an individual’s FP alter their ability to make a facial expression, but often people with FP will avoid making facial expressions to maintain more resting symmetry (Bradbury et al., 2006). Others can interpret this absence of expression as a lack of interest, which can lead these individuals to avoid interactions with the individual with FP (Bogart et al., 2012). In addition to conveying emotion, facial expression works to regulate the dynamics of a conversation (Ekman & Friesen, 1971). In FP, this is often not possible and people can be misinterpreted as being insincere (Bogart & Matsumoto, 2010).

Secondly, the Model of Maintenance of Shame Proneness in visible difference (Appendix N, Kent & Thompson, 2002) proposes that individuals with a visible difference withdraw from social contact as a way of coping. Qualitative research confirms that many people with FP cope by limiting social activities (Norris et al., 2019). People with FP report isolation due to a lack of understanding from others (medical professionals and friends/family) about the impact of FP (Hamlet et al., 2021).

Given the above and the fact that research which shows individuals with other stigmatised conditions (e.g. HIV) receive low levels of social support (Okonkwo et al., 2016), it is likely that individuals with FP will also receive less social support when compared to the general population.

Stigma

Goffman (1963) describes stigma as an attribute that differentiates an individual from others, putting them in a less desirable category. In the literature, stigma has been divided into two distinct constructs: public stigma and self-stigma (Corrigan et al., 2005; Corrigan & Watson, 2002). Public stigma refers to stereotypes endorsed by the majority. This could be perceived public stigma, defined as how an individual believes others would view or treat them (Pedersen & Paves, 2014), or enacted, defined as the experience of unfair treatment and discrimination by others (Gray, 2002). Self-stigma refers to a stigmatised individual internalising public stigma (Corrigan & Watson, 2002)

Research has consistently demonstrated that perceived public stigma is associated with poorer outcomes, including poorer mental health (Emmer et al., 2020), lower quality of life (Degnan et al., 2021), and poor adherence to medical treatment (Kamaradova et al., 2016). It is therefore important to research how to reduce the impact of stigma on stigmatised individuals.

The effects of stigma have been explored in a number of physical health conditions, such as Functional Neurological Disorder (Ko et al., 2022), chronic pain (Perugino et al., 2022) and epilepsy (Kwon et al., 2022). Individuals with FP experience significantly more stigma compared to the general population, in the form of discrimination, avoidance, and bullying (Bogart, 2020; Bogart et al., 2012). Higher rates of perceived stigma has also been found in children with facial difference, compared to controls (Masnari et al., 2012).

Stigma and Perceived Social Support

The elimination of public stigma requires global changes in views of stigmatised populations, which often happen very gradually (Corrigan et al., 2005). Whilst pushing to make these macro level societal changes, it is also important to identify ways of protecting stigmatised individuals from the negative effects of public stigma.

In recent research exploring predictors of psychosocial outcomes for individuals with any visible difference, perceived social support was found to predict positive outcomes (Zucchelli et al., 2023). Poor psychosocial outcomes predicated by perceived lack of social support included life disengagement, due to appearance related concerns, and intimacy distress (Zucchelli et al., 2023). The mechanisms of why this effect was observed were not tested.

Social support has been identified as an important coping resource when faced with any challenging situation (Sherbourne & Stewart, 1991). Within the stigma literature, emerging research shows that perceived social support buffers against the effect of anticipated public stigma on stress in people living with HIV (Earnshaw et al., 2015). Social Cognitive Processing Theory (SCP) aims to explain this buffering effect of social support on negative outcomes (Lepore, 2001). SCP draws on trauma theory which suggests that the active processing of a traumatic event leads to better psychological outcomes (Creamer et al., 1992). SCP hypothesises that receiving social support encourages individuals to express concerns over stressors, including stigmatising experiences or worries about future anticipated stigma. If the expression of the stressor is well received then this facilitates an individual's cognitive and emotional processing of the stressor, leading to better psychological adjustment. The theory also hypothesises that if concerns are received poorly then this can result in increased psychological distress (Major et al., 1997). We know from the FP research, social interactions for this population can be a negative experience (Bogart et al., 2012). As a relatively new area of research, there has been little research into the buffering effects of social support in physical health conditions other than HIV (Earnshaw et al., 2013), and none in FP.

Aims of Present Study

The current study explores the differences in perceived social support in the BP population, and the general population. The study also aims to explore whether perceived social support moderates the impact of perceived public stigma on negative outcomes (anxiety and depression) in people with BP. It is important to uncover factors which protect people with BP from the negative outcomes of experiencing stigma, as there is potential for this to be an intervention target. The research will focus on individuals with BP, as we know that acquired FP is associated with a higher rates of psychological distress compared to those born with FP (Bogart, 2020), and BP is the commonest type of FP.

Research Questions and Hypothesis

1. Do levels of perceived social support differ between individuals with BP and those in the general population?
    a. Hypothesis - Individuals with BP will report different levels of perceived social support than those in the general population.
2. Is perceived stigma correlated with distress in the BP group?
    a. Hypothesis - Perceived stigma is correlated with levels of distress in the BP group, as measured by the PHQ-8 and GAD-7.
3. Does perceived social support act as a moderator on the relationship between perceived public stigma and distress in individuals with BP?
    a. Hypothesis - Perceived social support moderates the relationship between perceived public stigma and distress (combined anxiety and depression) in individuals with BP.

Method

Design

The study employed a cross-sectional design. Participants completed an online survey using the Qualtrics XM Platform.

Patient and Public Involvement

Individuals with BP were consulted on the proposed design of the study and wording of the participant documents. The participant documents were altered in line with their feedback. They also advised on where to advertise the study, to optimise recruitment uptake.

Ethics

This study received approval from the Oxford University Central University Research Ethics Committee (CUREC; Reference number R86293/RE001; see Appendix O). All participants gave informed consent.

Participants

Individuals with BP

The study recruited adults (defined as being 18 years of age or older) who were experiencing BP at the time of the study. There was no minimum time for which a participant could have had BP. Participants were recruited during May 2023 via social media. Facial Palsy UK also advertised the study on their social media pages.

Inclusion criteria were as follows:

- Currently experiencing BP
- Able to speak good enough English to complete the questionnaires.
- Aged 18 or over.
- Able to consent to taking part in the study.
- Live in the UK

Exclusion criteria were as follows:

- Self-reported cognitive impairment, as this is likely to impact an individual’s ability to complete the questionnaires.
- Lack of English language as translation of the measures was not viable
- FP of any other type (e.g. congenital FP)

General Public Control Group

A comparable group was recruited from the general population via social media and word of mouth. Purpose sampling was adopted to ensure as close a match between samples as possible on key demographic variables (e.g. age, gender, ethnicity).

Inclusion criteria were as follows:

- Able to speak good enough English to complete the questionnaires.
- Aged 18 or over.
- Able to consent to taking part in the study.

Exclusion criteria were as follows:

- Any other type of visible difference (e.g. burn or scar)
- Cognitive impairment, as this is likely to impact an individual’s ability to complete the questionnaires.
- Lack of English language.

Power Analysis

G*Power software was used to conduct a power analysis to determine the sample size needed. In previous research, the effect size for difference in perceived social support between a reasonably equivalent facial burns population, and a healthy control group has been reported as *d* = 1.03 (Waqas et al., 2016). However, due to the differing populations and concerns about the quality of this research, we will be aiming for a more conservative medium effect size (*d* = 0.5). For the between group comparison, at least 64 participants were required in each group in order to detect a significant effect of *d* = 0.5 with 80% confidence (power = .80, alpha = .05)

Secondly, a power calculation was performed related to the moderation analysis. Previous research into the moderation effect of perceived social support has found a range of effect size from 0.085-0.13 (Earnshaw et al., 2015). Based off the smaller of the two effect sizes, and with power set at 80%, we would require a sample size of 95 in the BP group. Based off the larger effect size, and with power set at 80%, we would require a sample size of 62 in the BP group.

Given the above, we aimed to recruit a minimum of 64 people in both the BP and general population groups, with a maximum target of 95 per group.

Measures

Demographics

Demographic variables were taken from both groups, including age, gender, biological sex, and ethnicity.

Bell's Palsy Information

Participants in the BP group were asked for the month and year of onset of their BP, along with the perceived noticeability of their BP to themselves and to others (both on a 0-100 scale). They were also asked about their expectations of recovery (on a 0-100 scale).

Perceived Public Stigma Measure

Perceived public stigma was measured using the Perceived Stigmatisation Questionnaire (PSQ; Appendix P, used and reproduced with the author's permission; Lawrence et al., 2006). This is a 21-item measure of perceived public stigma experienced by people with a visible difference and has been shown to have a good construct validity (Lawrence et al., 2006). It asks participants how often they are treated in different ways. The results give an overall score from 0-5, as well as scores on three sub-scales: 1) Absence of friendly behaviour, 2) Staring/confused behaviour, and 3) Hostile behaviour. A higher score indicates greater levels of perceived stigma. The PSQ has been used in adult burn research (Lawrence et al., 2006). This was completed by the BP group only.

Perceived Social Support Measure

Perceived social support was measured using the Multidimensional Scale of Perceived Social Support (MSPSS; Appendix Q, used and reproduced with permission from the primary author; Zimet et al., 1988). This 12-item measure was completed by both the BP and control group. The results give an overall score of perceived social support from 0-7, as well as scores on three subscales: 1) Significant other, 2) Family and 3) Friends. A higher score indicates higher levels of perceived social support. The measure demonstrates good internal reliability and strong factorial validity (Zimet et al., 1990). The measure has been used in research into perceived social support in visible difference more generally (Zucchelli et al., 2023).

Distress

Overall distress was measured by combining a participant's scores on the Patient Health Questionnaire-8 (PHQ-8; Kroenke et al., 2009) and Generalised Anxiety Disorder Assessment-7 (GAD-7; Spitzer et al., 2006).

The PHQ-8 is an 8-item measure of depression symptoms. The results give an overall score from 0-24, with a higher score indicating more depression symptoms present. Compared to the PHQ-9 it misses out a risk question, making it more suitable for research (Kroenke et al., 2001). The PHQ-8 has been shown to have good criterion validity, construct validity and reliability (Kroenke et al., 2009; Shin et al., 2019).

The GAD-7 is a seven-item measure of anxiety symptoms. The measure gives an overall score from 0-21, with a higher score indicating more anxiety symptoms. It has demonstrated good reliability and good construct, criterion, factorial and procedural validity (Spitzer et al., 2006). It is recommended for use in the FP population clinically (Hotton et al., n.d.).

Procedure

The study was advertised on social media (Facebook, Twitter, and Instagram) by the primary author in May 2023. Facial Palsy UK also advertised the study on their website and social media pages. Participants were guided to a Qualtrics page from the study advertisements, the first page of which was the Participant Information Sheet (PIS). The researcher's email address was shared with participants, should they wish to ask any questions. Participants then completed a consent form at the end of the PIS, before completing the questionnaires. Participants had access to a debrief page upon completion of the survey.

Data Analysis

The data was downloaded from Qualtrics and analysed using SPSS (Version 29.0.1.0). A Chi-square and t-test was performed to explore differences between the BP and control group demographics. The ethnicity data violated the expected frequency assumptions so was transformed into 'white' and 'non-white' for the chi-square test. Extreme outliers of the questionnaire scores were identified using boxplots and subsequently removed from the data. SPSS deems responses as an extreme outlier if it lies above the upper quartile plus 3 times the inter-quartile range, or below the lower quartile minus 3 times the inter-quartile range. This led to four extreme outliers being identified and removed from the questionnaire responses, all were in the control group. Two respondents were removed from the MSPSS survey data, one from the PHQ-8 data, and one from the GAD-7 data.

Histograms, P-plots, and Q-plots were visually inspected to determine the normality of the data. They demonstrated a normal distribution of the data on the responses to the questionnaires. Therefore, parametric tests were used in the analysis.

A two-tailed independent T-test was used to assess whether the BP and control groups had significantly different levels of perceived social support, anxiety, and depression. The effect size used was Cohen's *d* (Cohen, 1988); effect sizes were described as small, 0.2, medium, 0.5, or large. 0.8 (Sullivan & Feinn, 2012). To explore perceived social support further, a mixed (2x3) ANOVA was run to determine whether levels of perceived social support differed between groups, by type of social support. A Pearson's correlation coefficient was calculated to explore the relationship between perceived stigma and distress outcomes, and to determine if any factors needed to be controlled for in the later moderation analysis. These were described as having low (0.1), moderate (0.3) or high (0.5) correlations (Cohen, 1988). A moderation analysis was undertaken using the PROCESS macro in SPSS to examine if perceived social support moderated the relationship between perceived stigma and distress outcomes. In preparing the data for moderation analysis, a composite score combining the PHQ-8 and GAD-7 was created using the method outlined by Song and colleagues (Song et al., 2013) to give an overall ‘distress’ score. A *z* score was calculated for each participant’s score on the two questionnaires. The z scores were then summed for each participant, to create a composite score of the two measures.

Results

Descriptive Statistics

Two-hundred and twenty-one people followed the Qualtrics link to the PIS, of whom 84% (185) completed the questionnaire and were included for analysis. Of these 185, 94 had BP and 91 were controls. Participant characteristics are shown in Table 3.1. The participants were mainly white and female. There was no association between group and gender, $\chi^2(1, N = 185) - .14$, $p = .71$, or ethnicity, $\chi^2(1, N = 185) = .46$, $p = .83$. There was not a significant difference between the mean ages of the two groups, $t(183) = .97$, $p = .33$.

Table 3.1

Participant Demographics

	BP Group (n=94)	Control Group (n=91)	Total (n=185)
Gender			
Female	82	81	163
Male	12	10	22
Other	0	0	0
Biological sex			
Female	82	81	163
Male	12	10	22
Other	0	0	0
Age			
Range	19-72	21-77	19-77
Mean (SD)	45.0 (13.5)	43.1 (13.6)	44.1 (13.5)
Ethnicity			
White	89	88	177
Mixed/multiple	0	0	0
Asian/Asian British	2	3	5
Black/African/Caribbean/Black British	2	0	2
Other	1	0	1

The BP specific characteristics are shown in Table 3.2.

Table 3.2

Participant Demographics

Bell's Palsy Characteristic	Range	*M*	*SD*
Time with BP (months)	0 – 587[1]	82.8	127.2
Noticeability of BP to you (0-100%)	5 – 100	83.1	23.2
Noticeability of BP to others (0-100%)	0 – 100	58.4	30.8
Expected recovery from BP (0-100%)	0 – 100	45.3	36.0

[1] 0 refers to people who have had BP for half a month or less, as this was rounded down to 0

A summary of the measures can be found in Table 3.3 below. All showed a very strong internal reliability.

Table 3.3

Cronbach's alpha (α), Means (M) and Standard Deviations (SD) for self-report scales.

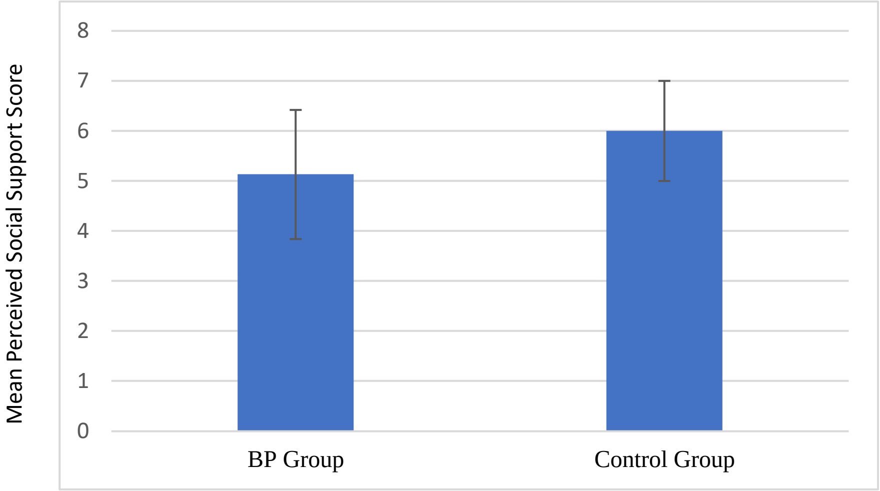
Measure	***α* for total sample**	***α* for BP group**	***M* for total sample**	***SD* for total sample**
Multidimensional Scale of Perceived Social Support	0.96	0.95	5.55	1.23
Perceived Stigmatisation Questionnaire	-	0.91	2.29	0.57
Patient Health Questionnaire-8	0.91	0.89	7.75	6.11
Generalised Anxiety Disorder Assessment-7	0.94	0.93	6.77	5.93

Research Question One - Do levels of perceived social support differ between individuals with BP and those in the general population?

People with BP (*M* = 5.1, *SD* = 1.3) reported significantly lower perceived social support than participants in the control group (*M* = 6.0, *SD* = 1.0), *t*(174) = -5.0, *p* <.001, *d* = -.73, indicating a medium effect size. The above T-test is visually presented in Figure 3.1 below.

Figure 3.1

Mean and standard deviation of MSPSS score



To explore the differences in perceived social support further, a mixed two-way 2 (BP or control group) x 3 (support type determined by the MSPSS subscales; significant other, family, friends) ANOVA was conducted. There was a significant main effect of group (BP vs control), $F(1,181) = 4197$, $p < .001$, $\eta_p^2 = .94$, with those with BP experiencing less perceived social support. There was also a main effect of support type (significant other vs family vs friend), $F(2,362) = 275$, $p < .001$, $\eta_p^2 = .47$. Post-hoc analysis, using pairwise comparisons with Bonferroni corrections, illustrated that across both groups, participants reported receiving significantly more social support from their significant other ($M = 5.31$, $SD = 1.71$), compared to their family ($M = 5.01$, $SD = 1.40$) and friends ($M = 3.80$, $SD = 0.98$), and significantly more social support from their family compared to their friends. The interaction was found not to be significant, $F(2, 362) = 2.02$, $p = .13$. This therefore shows that the levels of perceived social support are statistically similar between groups, across each type of social support.

Participants in the BP group reported significantly more depression symptoms on the PHQ-8 ($M = 11.4$, $SD = 6.2$) than the control group ($M = 3.9$, $SD = 2.8$), $t(131) = 10.7$, $p =$

<.001*,* *d* = 1.56, indicating a large effect size. Of note, the mean PHQ-8 score for the BP group is above the recommended clinical cut-off of ten (Kroenke et al., 2009) whilst the control group scores below cut off on average.

On the GAD-7, participants in the BP group (*M* = 9.96, *SD* = 6.3) reported significantly more symptoms of anxiety compared to the control group (*M* = 3.4, *SD* = 3.0), *t*(134) = 9.0, *p* = <.001*,* *d* = 1.31, indicating a large effect size. The mean GAD-7 score for the BP group is fractionally below the recommended clinical cut-off of 10, the control group average is well below the cut off (Spitzer et al., 2006).

Research Question Two - Is perceived stigma correlated with distress in the FP group?

A Pearson's correlation was performed exploring the relationship between perceived stigma and distress outcomes. As hypothesised, perceived stigma was significantly associated with distress (combined anxiety and depression) *r*(92) = .33, *p* < .001. This was a significant correlation with a medium effect size.

To better understand the relationships between measured variables, several correlation calculations were run with the data from the BP group. The significant findings are summarised in Appendix R. Age, length of time living with BP, noticeability of BP to self and others, and expected recovery were not associated with either measure of distress (the outcome of interest), and therefore not controlled for in the moderation analysis. Perhaps predictably, there was a significant correlation with a moderate effect size between time with BP and expected recovery, *r*(92) = -.43, *p* < .001, and a high correlation between noticeability of BP to self and others, *r*(92) = .72, *p* < .001. Interestingly, perceived social support was significantly associated with depression symptoms, *r*(92) = -.220, *p* = .033 (small effect size), but not anxiety symptoms.

Research Question Three - Does perceived social support act as a moderator on the relationship between perceived public stigma and distress in individuals with BP?

To address the third and final research question, a moderation analysis was performed using the PROCESS tool. The outcome variable was distress, the predictor variable was perceived stigma, and the potential moderator was perceived social support. The results are presented in table 3.4. Perceived social support was not an independent predictor of distress, $b = -.14$, 95% CI [-.46, .18], $t = -.86$, $p = .39$. Perceived stigma was an independent predictor of distress, $b = .96$, 95% CI [.23, 1.69], $t = 2.56$, $p = .012$. The interaction between perceived stigma and perceived social support was found not to be statistically significant, $b = -.13$, 95% CI [-.56, .31], $t = .58$, $p = .56$.

Table 3.4

Predictors of distress

	b	***SE B***	***t***	***p***
Constant	1.08	.20	5.45	.00
Perceived stigma	.96	.37	2.56	.012
Perceived social support	-.14	.16	-.86	.39
Interaction	-.13	.22	-.58	.56

Discussion

The current study found that people with BP report lower levels of perceived social support compared to the general population. This is in line with findings from other stigmatised health conditions such as HIV (Okonkwo et al., 2016). Qualitatively, people with FP consistently report difficulties in social interactions (Coulson et al., 2004; Hamlet et al., 2021). Many withdraw from social activities because of their FP (Norris et al., 2019). For others, their lack of facial expression, may be misinterpreted by others as a lack of interest so people withdraw from the conversation (Bogart et al., 2012). It could be a combination of these reasons that contributes to the observed effects within this study.

Using the PHQ-8 and GAD-7, the current study observed significantly higher levels of anxiety and depression symptoms within the BP population compared to the control group. This is in line with a number of previous studies finding the same effect with different measures of anxiety and depression (e.g. Pouwels et al., 2016; Walker et al., 2012). To the author's knowledge, this is the first study to compare anxiety and depression levels in the BP and general population using the PHQ-8 and GAD-7. Both showed a very strong internal consistency within the BP sample. Of note, the mean PHQ-8 score fell above the recommended clinical cut off for depression and the GAD-7 mean score was very close to this. Of note, perceived social support was significantly associated with depression, but not anxiety. The authors hypothesise that this could be a bidirectional relationship, with people with BP and depression symptoms withdrawing from social activities due to low mood (Girard et al., 2014), but also we know that poor perceived social support predicts depression (de la Vega et al., 2019).

As with other stigmatised physical health conditions, such as HIV (Ow & Lee, 2015), perceived stigma was found to be associated with anxiety and depression. Contrary to our hypothesis, perceived social support did not moderate this relationship. The current study focused on perceived social support as a potential moderator due to emerging literature in visible difference research regarding social support (e.g. Zucchelli et al., 2023). However, the Mediated-Moderation Model of Self-Compassion and Stigma (Wong et al., 2019) describes social support as one element of self-compassion, a construct believed to buffer the relationship between stigma and psychological outcomes. Other aspects such as 'intrinsic self-affirmation' could be important in moderating the effect of stigma on distress. This aspect refers to losing sight of valued identities as they become overshadowed by the stigmatised identity. In acquired FP, people often feel as though they have lost the sense of their old identity (Hamlet et al., 2021) so this element could be relevant here. Therefore, intrinsic self-affirmation as an element of self-compassion may be worth exploring as a potential moderator in future research.

Clinical Implications

It has been well established that objective severity of FP does not predict distress (e.g. Díaz-Aristizabal et al., 2019). The current study adds to this by demonstrating that subjective noticeability (to yourself and others), and expected recovery were also not associated with distress. This is in line with previous research which shows that length of time with BP is not associated with anxiety (Walker et al., 2012) or depression (Fu et al., 2011). The above is important to bear in mind clinically. It has emphasised the fact that healthcare professionals cannot make assumptions based on characteristics of an individual's BP about who will experience the most distress. A comprehensive assessment and formulation should guide any psychological input. The elevated levels of distress in the BP group demonstrates the need for Psychologists within FP MDT.

Limitations and future directions

The sample for this study was largely white, females. While this is comparable to other studies in visible difference (Zucchelli et al., 2023), it limits the generalisability of the findings. Exploring the experiences of people with FP from different ethnicities is an important area for future research to focus on. Research into the stigma of mental illness has found that perceived stigma can differ depending on an individual's ethnicity (Eylem et al., 2020). In an individual who experiences stigma for any reason, ‘intersectional discrimination’ can occur. This is when a person has two or more attributes which interact to produce a distinct form of stigma (Atrey, 2018). A qualitative study looking into Alopecia Areata, a different type of visible difference, described a compounding effect of a stigmatised appearance based on both the colour of an individual’s skin on top of their visible difference (Zucchelli et al., 2022). It is therefore important to be mindful of the experience of people who may be experiencing intersectional discrimination. There is no research into the experience of people from different ethnicities in the FP population and, due to the reasonable homogeneity of the sample in the current study, we were not able to explore this.

In hindsight, it may have been better to use a visible difference specific measure of distress. For example, the FACE-Q could have been employed here (Klassen et al., 2010). Although it would not have allowed comparison between the BP and control group, were social support to play a moderating role between stigma and distress, there is a chance this may have been more detectable through the use of an outcome measure more specific to BP.

There were also limitations to the study design. Causal conclusions cannot be drawn from exploratory, cross-sectional data. Therefore, longitudinal and experimental designs are needed to establish the directions of the relationships discussed above, particularly, whether changes in perceptions of stigma or social support lead to changes in anxiety and/or depression.

Perceived stigma was found to independently predict distress. While this highlights the need for campaigns to improve awareness and reduce public stigma around FP, it also opens the door to future research into interventions to reduce the impact of stigma on an individual. Peer-led interventions and peer support groups have been shown to alleviate stress caused by stigmatising experiences in other stigmatised groups (J. Sun et al., 2022). A single session peer support group has been piloted in children with FP and their parents and shows preliminary evidence of being a successful approach (Hotton et al., 2019). Similar groups have been shown to have positive outcomes for adults with congenital FP (Bogart & Hemmesch, 2016). This study opens the door for such support groups to be trialled with people with BP.

Conclusion

This study has found that compared to controls, people with BP have significantly lower perceived social support, and significantly higher rates of depression and anxiety symptoms. Subjective noticeability of BP (to self and others), length of time with BP and expected recovery were not associated with distress, therefore reminding clinicians not to assume who may be struggling psychologically. Perceived stigma was found to predict distress (depression and anxiety symptoms), but this relationship was not moderated by perceived social support. Other potential moderators within the Mediated-Moderation Model of Self-Compassion and Stigma should be explored in future research.

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Executive Summary

Why did we do this study?

Bell’s Palsy (BP) is a type of acquired Facial Palsy (FP). With weakened facial muscles, social interactions can be difficult for people with BP. Due to feeling anxious or self-conscious, people with BP can withdraw from social activities. Also given the difficulties that people with BP have in making different facial expressions, other around them can mis-interpret their lack of facial expression as meaning they are uninterested or disingenuous. We know from previous research that social support helps people to cope with difficult life events. Therefore, our first research question investigated whether levels of social support are different in people with BP compared to the general population.

Previous research has demonstrated that people with FP can experience stigma. In other populations, experiencing stigma has found to be associated with poorer outcomes. Therefore, our second research question investigated whether stigma was associated with distress (anxiety and depression).

If stigma is associated with distress, we need to explore how we can help protect individuals with BP from the negative impacts of stigma. As above, social support helps individuals to cope through difficult time. We therefore tested whether social support from others buffers against the negative impacts of stigma.

How was the study carried out?

People with BP acted as advisors in the design of the study, and any changes they recommended were incorporated into the final design.

Adults with BP were recruited through social media. The charity Facial Palsy UK also posted about the study on their website. To be able to compare people with BP to the general population, a control group was also recruited via social media.

Both groups took part in an online survey and completed measures of social support, anxiety, and depression. The people with BP also completed a measure of stigma and filled out some information about their BP.

What did the study find?

One-hundred and eighty-five participants took part in the project. Ninety-four of these had BP and ninety-one were controls. Across both groups, most participants were white females.

As we predicted, we found that people with BP reported that they have less social support compared to controls. We also found out that people with BP had significantly more anxiety and depression symptoms compared to the control group.

We found that the more people with BP reported feeling stigmatised, the more distress (anxiety and depression) they felt. Interestingly, we found that the characteristics of an individual's BP was not associated with distress. This included measures of noticeability of the BP to self and others, length of time with BP and expected recovery.

Finally, we found that social support did not buffer against the negative impacts of stigma as we had first thought.

What did we conclude?

We concluded that the continued psychological support of people with BP is crucial, given then show higher levels of anxiety and depression, and lower levels of social support compared to the general population. Given the fact that features of an individual's BP is not associated with their distress levels, we remind clinicians that they cannot assume who will (or won't) be more psychologically effected. As social support was found not to buffer against the negative impacts of stigma, we recommend further research is conducted to explore other potentially buffering constructs.

Connecting Narrative

My papers share a common theme: health. It is an area I have always been interested in, having spent a lot of my childhood visiting family members in hospital. As I look forward to qualified life, I have known for most of my training journey that I see myself working as a Psychologist in a Health setting. Therefore, choosing three projects which each, in their own way, explore how a Psychologist can better support those with a physical health diagnosis made total sense for me. I had enjoyed working as a Research Assistant at the UCL Great Ormond Street Institute of Child Health prior to training, and this had given me a strong foundational knowledge about how to conduct research within different health fields. My reflections on the process of conducting each research project are detailed below.

Systematic Review of the Literature

I first came across non-epileptic attack disorder (NEAD) when working as an Assistant Psychologist in an Epilepsy specialist centre, working with some of the world leading NEAD experts. I found it a fascinating area to work in. Being part of a team which often diagnosed people with NEAD, I saw first-hand how people received that diagnosis very differently. For some it was a relief, for some it was a shock, for some they were angered by it. It was well known in research and clinical practice that people with NEAD often find it difficult to engage in therapy. Knowing how successful therapy could be, I wanted to understand this further, which led me to designing this systematic review.

Having never done a systematic review before, I learned a lot as I moved through the process. The hardest part of the review was defining the search terms. There are so many interchangeable terms for ‘NEAD’ and ‘therapy’ that it took a lot of refining and talking to experts to make sure I had all basis covered. I found asking the librarians for support particularly helpful.

Finally, I completed my review first out of my three projects. Working through the quality rating tool for each study helped me to refine my service improvement project and theory driven research project. I knew what a ‘high quality’ study looked like and was able to keep this in mind when designing and writing up my other two projects.

Service Improvement Project

I spoke to a number of different physical health teams when it came to deciding on my service improvement project. In my discussions with the Craniofacial team, I was really struck by the responsibility that is put onto parents in making the decision about whether to pursue corrective surgery for Craniosynostosis. To weigh up the decision of putting a seemingly healthy child through skull remodelling surgery seemed like an impossible decision, and I could see how parents may rely on a psychologist to support them through this process. Helping to make this journey as smooth as possible for parents was an opportunity that really appealed.

The most challenging part of this project was staying under the word limit in the write up. Fitting the analysis of 28 qualitative surveys into 5,000 words was not an easy task. I found it helpful to be reminded by a supervisor what within the write-up, I was telling a story to a reader and to focus on the take-home messages, rather than trying to recount every participant’s experiences. I found it particularly rewarding to read through the parent surveys as some were so incredibly grateful to each member of the multi-disciplinary team. A lot mentioned specific MDT members by name and recounted memorable instances where their support had been invaluable. By the time I was analysing the data, I was on placement with the Cleft, Craniofacial and Facial Palsy teams so knew a lot of the named healthcare professionals.

Theory Driven Research Project

In December of my final year, I made the difficult decision to re-start my project as I was experiencing significant recruitment difficulties with my original idea. I was fortunate enough to be in a placement I was really enjoying, working in a specialist Facial Palsy (FP) team. In reading around at the beginning of my placement, I was struck by the limited research in this area compared to other conditions known to be associated with stigma. Working day-to-day on placement listening to people recount experiencing stigma and the understandable impact this had on their mental health, I began to think about how this could form a research question. Through the invaluable support of my supervisors, I designed a project exploring an element of this. I was very aware that while I was under significant time pressure, I did not want to rush the process of designing my second project. I think this is a mistake I fell into first time around and didn't think through the realities of the project well enough, instead rushing it through the PAS process. It was a fine line to balance.

A stressful six month ensued trying to push this project through on time. I found working with people with FP, and within a team so passionate about helping people with FP, was so helpful to keeping up my motivation. Reflecting on my second project now, I realise how different it was to my first-time round. I thought that the time that I invested in my first project idea had gone to waste, but the process taught me so much. When it came to designing a second project, applying for ethical approval, and recruiting, I was so much more confident. I knew the process, felt much more confident in my abilities, and knew when to ask for help from supervisors. Taking this forward into qualified life, I can see how strong my research skills now are. I look forward to taking this further, but without such a tight deadline to aim for!

Acknowledgements

I am incredibly grateful to the patients and healthcare professionals who took part in the research described in this thesis. Thank you for trusting me with your stories and experiences.

I would like to thank my research supervisors, Sam Lloyd-White, Katie Hamilton, Susannah Jenner, Fiona McFarlane and Ceara Moore. Your support and guidance over the last three years has been so appreciated. A special thank you to Matt Hotton, whose encouragement and belief in me over the last year has got me across this finish line in one piece. Thank you to my course tutors, Sarah Lack and Matthew Knight. You have both been pivotal in helping me to look after myself during the rollercoaster of the doctorate. Thank you to my personal tutor, Camilla Holden, for always being on the end of the telephone and offering such helpful advice.

A big thank you to the ‘Covid cohort’ of 2020. Starting this journey in the height of the pandemic was a daunting prospect but you all made it so enjoyable. Thank you especially to Chiara, Joe, Bethan, and Josh. I couldn’t have hoped for better friends to see me through the last three years. We have laughed, cried, danced, ranted, and everything in between. Chiara, thank you for the cups of tea when I needed them most.

The most heartfelt thank you to everyone in my personal life who have been there through the highs and lows of this doctorate. To H, Nannie, Dad, Gail, Clara, Simon, Charl, Sarah, Dave, and Izzy. Thank you for keeping me grounded, always being interested in the work I am doing, and most importantly for helping me to switch off! George, thank you for being my biggest cheerleader. Thank you for always encouraging me to believe in myself and for your unwavering patience during the ups and downs of the last three years.

Finally, to my mum. Thank you for fostering my curiosity from such a young age and encouraging me to work hard towards my dreams. Thank you for suggesting a career in Clinical Psychology.

Appendix A

SRL: Author guidelines for Epilepsy & Behaviour

INTRODUCTION

Epilepsy & Behavior has been, and still is, the fastest-growing international journal since its launch in 2000.

Epilepsy & Behavior is uniquely devoted to the rapid dissemination of the most current information available on the behavioral aspects of seizures and epilepsy.

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- Journal policies detailed in this guide have been reviewed
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This journal encourages you to cite underlying or relevant datasets in your manuscript by citing them in your text and including a data reference in your Reference List. Data references should include the following elements: author name(s), dataset title, data repository, version (where available), year, and global persistent identifier. Add [dataset] immediately before the reference so we can properly identify it as a data reference. The [dataset] identifier will not appear in your published article.

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Examples:
Reference to a journal publication:

[1] Van der Geer J, Hanraads JAJ, Lupton RA. The art of writing a scientific article. J Sci Commun 2010;163:51–9. https://doi.org/10.1016/j.Sc.2010.00372.
Reference to a journal publication with an article number:
[2] Van der Geer J, Hanraads JAJ, Lupton RA. The art of writing a scientific article. Heliyon. 2018;19:e00205. https://doi.org/10.1016/j.heliyon.2018.e00205

Reference to a book:
[3] Strunk Jr W, White EB. The elements of style. 4th ed. New York: Longman; 2000.
Reference to a chapter in an edited book:
[4] Mettam GR, Adams LB. How to prepare an electronic version of your article. In: Jones BS, Smith RZ, editors. Introduction to the electronic age, New York: E-Publishing Inc; 2009, p. 281–304. Reference to a website:
[5] Cancer Research UK. Cancer statistics reports for the UK, http://www.cancerresearchuk.org/ aboutcancer/statistics/cancerstatsreport/; 2003 [accessed 13 March 2003].
Reference to a dataset:
[dataset] [6] Oguro M, Imahiro S, Saito S, Nakashizuka T. Mortality data for Japanese oak wilt disease and surrounding forest compositions, Mendeley Data, v1; 2015. https://doi.org/10.17632/ xwj98nb39r.1.
Note shortened form for last page number. e.g., 51–9, and that for more than 6 authors the first 6 should be listed followed by 'et al.' For further details you are referred to 'Uniform Requirements for Manuscripts submitted to Biomedical Journals' (J Am Med Assoc 1997;277:927–34) (see also Samples of Formatted References).

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Appendix B

Permission to re-produce World Health Organisation Multidimensional Adherence Model

Email received on 4th April 2023

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12. Headings. Paragraph headings in this Agreement are for reference only.

13. Dispute resolution. Any dispute relating to the interpretation or application of this Agreement shall, unless amicably settled, be subject to conciliation. In the event of failure of the latter, the dispute shall be settled by arbitration. The arbitration shall be conducted in accordance with the modalities to be agreed upon by the parties or, in the absence of agreement, with the rules of arbitration of the International Chamber of Commerce. The parties shall accept the arbitral award as final.

14. Privileges and immunities. Nothing in or relating to this Agreement shall be deemed a waiver of any of the privileges and immunities enjoyed by WHO under national or international law and/or as submitting WHO to any national court jurisdiction.

Appendix C

SRL Search Terms

(“non-epileptic attack disorder” OR NEAD OR “non epileptic attack disorder” OR PNEA OR “psychogenic non-epileptic attack*” OR “psychogenic non epileptic attack*” OR “non-epileptic attack*” OR “non epileptic attack*” OR “non-epileptic seizure*” OR “non epileptic seizure*” OR “non epileptic fit*” OR “non-epileptic fit*” OR “non epileptic convulsion*” OR “non- epileptic convulsion*” OR “psychogenic seizure*” OR “psychogenic fit*” OR “psychogenic convulsion*” OR “psychogenic non-epileptic event*” OR “psychogenic non epileptic event*” OR “pseudo-seizure*” OR “pseudo seizure*” OR “pseudo-fit*” OR “pseudo fit*” OR “pseudo-convulsion*” OR “pseudo convulsion*” OR “dissociative seizure*” OR “dissociative fit*” OR “dissociative convulsion*” OR “Psychogenic non-epileptic seizure*” OR PNES OR “Non epileptic paroxysmal event*” OR NEPE OR “conversion disorder”)

AND

(engage* OR adher* OR refus* OR retention OR retain OR “non-adherence” OR “Non adherence” OR “Non complian*” OR “Non-complian*” OR “drop-out*” OR “Drop out*” OR “non-completion” OR “non completion” OR barrier* OR “dis-engage*” OR disengage* OR facilitate* OR uptake OR “Health care barrier*” OR Complian* OR Adher* OR Participat* OR “No show*” OR “No-show*” OR Attendance)

AND

(“psychological therap*” OR therap* OR CBT OR “Cognitive Behavio* Therap*” OR psychotherapy* OR Psychoeducation OR EMDR OR “Eye Movement Desensitization and Reprocessing” OR “Psychological Intervention*” OR “Psychological Treatment*” OR “Acceptance and Commitment Therap*” OR ACT OR Psychotherap* OR psychodynamic)

Appendix D

Mixed Methods Appraisal Tool (Hong et al., 2018)

Category of study designs	**Methodological quality criteria**	**Responses**		
		Yes	No	Can’t tell
Screening questions (for all types)	S1. Are there clear research questions?			
	S2. Do the collected data allow to address the research questions?			
	Further appraisal may not be feasible or appropriate when the answer is ‘No’ or ‘Can’t tell’ to one or both screening questions.			
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?			
	1.2. Are the qualitative data collection methods adequate to address the research question?			
	1.3. Are the findings adequately derived from the data?			
	1.4. Is the interpretation of results sufficiently substantiated by data?			
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?			
2. Quantitative randomised controlled trials	2.1. Is randomization appropriately performed?			
	2.2. Are the groups comparable at baseline?			
	2.3. Are there complete outcome data?			
	2.4. Are outcome assessors blinded to the intervention provided?			
	2.5 Did the participants adhere to the assigned intervention?			
3. Quantitative non-randomised	3.1. Are the participants representative of the target population?			
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?			
	3.3. Are there complete outcome data?			
	3.4. Are the confounders accounted for in the design and analysis?			
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?			
4. Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?			
	4.2. Is the sample representative of the target population?			
	4.3 Are the measurements appropriate?			

	4.4 Is the risk of non-response bias low? 4.5. Is the statistical analysis appropriate to answer the research question?
5. Mixed methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question? 5.2 Are the different components of the study effectively integrated to ask the research question? 5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted? 5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed? 5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?

Appendix E

SIP: Author guidelines for submission

The Cleft Palate and Craniofacial Journal

1. What do we publish?

1.1 Aims & Scope

Before submitting your manuscript to *CPCJ*, please ensure you have read the Aims & Scope. *CPCJ* publishes manuscripts of the highest scientific quality on all topics related to orofacial clefts and other craniofacial anomalies to advance the global education of scientists and clinicians.

Only manuscripts of sufficient quality that meet the aims and scope of *The Cleft Palate Craniofacial Journal (CPCJ)* will be reviewed. *CPCJ* is directed to a multidisciplinary readership of clinicians and scientists interested in craniofacial anomalies, including cleft lip and cleft palate.

1.2 Article Types

Research

Article Type	Description	Requirements

Original Research	Reports of original clinical or basic science data pertaining to prevalence, causes, mechanisms, diagnosis, course, treatment, and prevention, including systematic reviews and meta-analysis that represent a new contribution to the field.
	Traditional, narrative reviews should also be considered in this category.
		- 7.000 words: Body of the manuscript (excluding abstract, references and figure captions)
		- <250-word structured abstract* using the below headings:
		  - Objective
		  - Design
		  - Setting
		  - Patients, Participants
		  - Interventions
		  - Main Outcome Measure(s)
		  - Results
		  - Conclusions
		- Minimum of 3 keywords
		- ≤6 figures and tables, combined
		- For systematic reviews and meta-analyses, please follow the PRISMA checklist and include the checklist in your list of files upon submission
		- For Clinical Trials, please include the CONSORT flow chart as a cited figure and the completed CONSORT checklist

		should be uploaded with your submission *Narrative review articles may have an unstructured abstract

1.3 Writing your paper

The SAGE Author Gateway has some general advice and on how to get published, plus links to further resources.

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2. Editorial policies

2.1 Peer review policy

When a manuscript is submitted, CPCJ editorial staff perform and initial evaluation according to the following criteria: material is original and timely, writing is clear, study methods are appropriate, data are valid, conclusions are reasonable and supported by the data, information is important, and topic has general interest to readers of this journal. From these basic criteria, the editors assess a paper's suitability for publication. Suitable manuscripts are sent to expert consultants for peer review. Manuscripts deemed unsuitable for publication are rejected promptly.

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2. Drafted the article or revised it critically for important intellectual content,
3. Approved the version to be published,
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2.8 Reporting guidelines

The relevant EQUATOR Network reporting guidelines should be followed depending on the type of study. For example, all randomized controlled trials submitted for publication should include a completed CONSORT flow chart as a cited figure and the completed CONSORT checklist should be uploaded with your submission as a supplementary file. Systematic reviews and meta-analyses should include the completed PRISMA flow chart as a cited figure and the completed PRISMA checklist should be uploaded with your submission as a supplementary file. The EQUATOR wizard can help you identify the appropriate guideline.

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2.9 Research data

At SAGE we are committed to facilitating openness, transparency and reproducibility of research. Where relevant, *CPCJ* requests all authors submit any primary data used in their research articles alongside their article submissions to be published in the online version of the journal or provide detailed information in their articles on how the data can be obtained. This information should include links to third-party data repositories or detailed contact information for third-party data sources. Data available only on an author-maintained website will need to be loaded onto either the journal’s platform or a third-party platform to ensure continuing accessibility.

Examples of data types include but are not limited to statistical data files, replication code, text files, audio files, images, videos, appendices, and additional charts and graphs necessary to understand the original research. The editor may consider limited embargoes on proprietary data. The editor can also grant exceptions for data that cannot legally or ethically be released. All data submitted should comply with Institutional or Ethical Review Board requirements and applicable government regulations. Authors should also follow data citation principles.

For more information please visit the SAGE Author Gateway, which includes information about SAGE's partnership with the data repository Figshare.

Author have the option of including a data availability statement during the submission process.

2.10 Cover letter

Cover letters are required when addressing topics such as IRB exceptions (Sect 2.6) or anything related to third-party submissions (Sect 4.6.1).

If none of these situations apply, then cover letters are optional. However, CPCJ encourages cover letters if authors have special information they wish to declare or disclose. For example, authors may wish to outline why their research is innovative or novel.

The manuscript submission system requires that cover letters be submitted as Microsoft Word documents.

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3. Publishing Policies

3.1 Publication ethics

SAGE is committed to upholding the integrity of the academic record. We encourage authors to refer to the Committee on Publication Ethics' International Standards for Authors and view the Publication Ethics page on the SAGE Author Gateway.

3.1.1 Plagiarism

The Cleft Palate Craniofacial Journal (CPCJ) and SAGE take issues of copyright infringement, plagiarism or other breaches of best practice in publication very seriously. We seek to protect the rights of our authors and we always investigate claims of plagiarism or misuse of published articles. Equally, we seek to protect the reputation of the journal against malpractice. Submitted articles may be checked with duplication-checking software. Where an article, for example, is found to have plagiarised other work or included third-party copyright material without permission or with insufficient acknowledgement, or where the authorship of the article is contested, we reserve the right to take action including, but not limited to: publishing an erratum or corrigendum (correction); retracting the article; taking up the matter with the head of department or dean of the author's institution and/or relevant academic bodies or societies; or taking appropriate legal action.

3.1.2 Prior publication

If material has been previously published it is not generally acceptable for publication in a SAGE journal. However, there are certain circumstances where previously published material can be considered for publication. Please refer to the guidance on the SAGE Author Gateway or if in doubt, contact the Editor at the address given below.

Please note as part of the submission process you will be required to warrant that you are submitting your original work, that you have the rights, that you have obtained and can supply all necessary permissions for the reproduction of any copyright works not owned by you, that you are submitting the work for first publication in the Journal, and that it is not being considered for publication elsewhere and has not already been published elsewhere. \

Note that the Journal may accept submissions of papers that have been posted on pre-print servers; include the DOI for the preprint in the designated field during the submission process. Authors should not post an updated version of their paper on the preprint server while it is being peer reviewed for possible publication in the journal. If the article is accepted for publication, the author may re-use their work according to the Journal's author archiving policy. If your paper is accepted, you must include a link on your preprint to the final version of your paper.

3.2 Contributor's publishing agreement

Before publication, SAGE requires the author as the rights holder to sign a Journal Contributor's Publishing Agreement. SAGE's Journal Contributor's Publishing Agreement is an exclusive licence agreement which means that the author retains copyright in the work but grants SAGE the sole and exclusive right and licence to publish for the full legal term of copyright. Exceptions may exist where an assignment of copyright is required or preferred by a proprietor other than SAGE. In this case copyright in the work will be assigned from the author to the society. For more information please visit the SAGE Author Gateway.

3.3 Open access and author archiving

CPCJ offers optional open access publishing via the SAGE Choice program. For more information on Open Access publishing options at SAGE please visit SAGE Open Access. For more information on Open Access publishing options at SAGE and pricing, as well as possible discounts or waivers, please visit SAGE Open Access.

4. Preparing your manuscript for submission

CPCJ is hosted on SAGE Track, a web based online submission and peer review system powered by ScholarOne™ Manuscripts. Visit https://mc.manuscriptcentral.com/cpcj to login and submit your article online.

IMPORTANT: Please check whether you already have an account in the system before trying to create a new one. If you have reviewed or authored for the journal in the past year it is likely that you will have had an account created. For further guidance on submitting your manuscript online, please visit ScholarOne.

Before entering the online manuscript submission system, please be sure the following elements are on hand:

- Contact details for all authors
- Funding disclosure details (when applicable – see Sect 2.4)
- Main manuscript files, including a separate title page (required) and separate tables and figures (if included)
- Any supplemental files (optional)
- Cover Letter (see Sect. 2.10 for when this is required)
- Completed PRISMA checklist (for systematic reviews and meta-analyses)
- Completed CONSORT checklist (for clinical trials)
- Twitter handles for authors and a drafted tweet of no more than 280 characters (optional)
- A data availability statement (optional, unless required by funder or institution)

4.1 File format

The preferred format for your manuscript is Word. LaTeX files are also accepted. Word and (La)Tex templates are available on the Manuscript Submission Guidelines page of our Author Gateway. Please ensure your manuscript is in either Word or LaTeX otherwise it may be sent back to you.

4.2 Make your article discoverable

When writing up your paper, think about how you can make it discoverable. The title, keywords and abstract are key to ensuring readers find your article through search engines such as Google. For information and guidance on how best to title your article, write your abstract and select your keywords, have a look at this page on the Gateway: How to Help Readers Find Your Article Online

4.3 Identifiable information

CPCJ uses double-anonymized peer review, and thus authors are required to submit:

1. A **version of the manuscript** which has any information that compromises the anonymity of the author(s) removed. This version **will** be sent to the peer reviewers.
2. A **separate title page** which includes any potentially identifying material. This **will not** be sent to the peer reviewers.

See this page for detailed guidance on making an anonymous submission.

4.4 Use of “Patient-First” Language

Please be sure you are using patient-first language in your entire manuscript (e.g., use "patients with CLP" instead of "CLP patients"; or "patients with 22q11.2 DS" instead of 22q11.2DS patients").

4.5 Avoiding Priority Claims

Manuscripts should avoid priority claims such as "this is the first study to...", "this is the largest study", etc. even when qualified by statements like "to our knowledge..."

4.6 Manuscript files to be uploaded

These include: Title Page (required); Manuscript (required); Tables (optional); Figures (optional); Supplemental Materials (optional).

4.6.1 Title Page

The Title Page (submitted separately from the manuscript) must include (in the following order):

- Title (maximum 20 words); should be informative, relevant, and concise
- Author names with *no more than* three highest attained degrees, in the order that they will appear in print
- Institutional affiliation for each author. The affiliation listed should be the institution where the research was conducted. If an author has moved to a new institution since completing the research, the new affiliation can be included in a manuscript note at the end of the paper.
- Name, address, telephone number, fax number, and email address of the corresponding author, who will receive all editorial communication and reprint requests
- Declaration of conflicting interest statement. Authors must disclose any relevant conflict of interest, including direct or indirect financial interests they may have in the materials or subject matter dealt with in the manuscript. If no conflict exists, please state that '*The Author(s) declare(s) that there is no conflict of interest*'.
- Any Acknowledgements to be included in the manuscript (see details below)
- If applicable, statement that manuscript was presented at a professional meeting, including the name, date, and location of the meeting
- Running title (less than 8 words)

Acknowledgments

All contributors who do not meet the criteria for authorship should be listed in an Acknowledgements section. Examples of those who might be acknowledged include a person who provided purely technical help, or a department chair who provided only general support.

If professional writing assistance was provided (e.g., from a specialist communications company) this should be included in the Acknowledgements section. Authors must disclose any writing assistance – including the individual's name, company and level of input – and identify the entity that paid for this assistance.

When an individual who is not listed as an author submits a manuscript on behalf of the author(s), a statement must be included in the Acknowledgements section of the manuscript and in the accompanying cover letter. The statement must confirm that the listed authors have authorized the submission of their manuscript via third party and approved any statements or declarations, e.g., conflicting interests, funding, etc.

Where appropriate, SAGE reserves the right to deny consideration to manuscripts submitted by a third party rather than by the authors themselves.

Do not include funding information in the Acknowledgements. Authors will be asked to disclose any sources of funding during submission.

To ensure that the article is anonymized, please do not include author names or affiliations, or any other identifying information in any portion of the manuscript other than this Title Page.

A formatted title page example can be found here for reference.

4.6.2 Manuscript

Word counts and specific formatting requirements for different article types are further described in Section 1.2. A formatted manuscript example can be found here for reference.

Page 1: Title The first page of the manuscript text file should include only the title used on the Title Page (above).

Page 2: Abstract Original articles and Ideas and Innovations articles should include a structured abstract of no longer than 250 words with the following headings and information, as applicable.

Structured Abstract:

- *Objective:* State the main question or objective of the study and the major hypothesis tested, if any.
- *Design:* Describe the design of the study indicating, as appropriate, use of randomization, anonymization, criterion standards for diagnostic tests, temporal direction (retrospective or prospective), etc.
- *Setting:* Indicate the study setting, including the level of clinical care (for example, primary or tertiary; private practice or institutional).
- *Patients, Participants:* State selection procedures, entry criteria, and numbers of participants entering and finishing the study.
- *Interventions:* Describe the essential features of any intervention, including the methods and duration of administration.
- *Main Outcome Measure(s):* The primary study outcome measures should be indicated as planned before data collection began. If the hypothesis being reported was formulated during or after data collection, this fact should be clearly stated.
- *Results:* Describe measurements that are not evident from the nature of the main results and indicate any anonymization. If possible, the results should be accompanied by confidence intervals (most often the 95% interval) and the exact level of statistical significance. For comparative studies, confidence intervals should relate to the differences between groups. Absolute values should be indicated when risk changes or effect sizes are given.
- *Conclusions:* State only those conclusions of the study that are directly supported by data, along with their clinical application (avoiding overgeneralization) and/or whether additional study is required before the information should be used in clinical settings. Equal emphasis must be given to positive and negative findings of equal scientific merit.

(Reproduced with permission from: Haynes RB et al. More informative abstracts revisited. *Ann Intern Med.* 1990;113:69–76).

Data-based Brief Communications articles should include a structured abstract of no longer than 150 words with the following headings: Objective, Design, Setting, Patients/Participants, Interventions, Main Outcome Measure(s), Results, Conclusions.

Non-data-based Brief Communications, Perspective articles, and Ethics/Health Policy reports should include an unstructured abstract of no longer than 100 words.

Case/Clinical reports should include an unstructured abstract of no longer than 100 words, describing the objective, essential features and uniqueness of the case being presented, and conclusions.

What I (We) Do articles should include a 50–75-word structured abstract with the following format: background (what is the issue/problem), solution, what I/we did that is new.

Narrative reviews should include an unstructured abstract of no longer than 250 words.

Letters to the Editor and Editorials do not require abstracts.

A note about Key Words: *Please do not include a list of Key Words in the manuscript. During manuscript submission, authors will be asked to select Key Words from a list of curated terms. The minimum number of Key Words required is three*.

Page 3: Body of Manuscript. Where applicable, divide the body of the manuscript into the Introduction, Methods, Results, Conclusion, References, and Figure Legends (if figures are included).

Additional details on the References and Figure Legends are included below.

If accepted, a Declaration of Conflicting Interests statement and a Funding Disclosure statement will be added to the manuscript during production. If any Acknowledgments were included on the title page, this text will also be added to the manuscript during production and will appear just before the references.

The *CPCJ* follows guidelines published in the *American Medical Association Manual of Style*.

- Manuscripts should be typed double-spaced with 1” margins, left justified, and use a standard 12-point font.
- Pages should be numbered consecutively in the upper right-hand corner.
- Do not print a running title.
- Turn off the word processing program’s hyphenation feature and ‘‘smart quotes’’ feature before typing.
- Headings must be used to designate the major divisions of the manuscript. Up to three levels of headings may be used.

Statistics

If a statistical analysis is conducted, explanation of the methods used must precede the Results section in the manuscript. Unusual or complex analysis methods should be referenced.

Units of Measure/ Abbreviations

The metric system is preferred for expressing units of measure. Abbreviations may be used for terms. The full term for each abbreviation should appear at its first use in the text, unless the abbreviation is a standard unit of measure. Abbreviations used in a table must be explained in a footnote below the table. For a list of standard abbreviations, consult the Council of Biology Editors Style Guide (available from the Council of Science Editors, 9650 Rockville Pike, Bethesda, MD 20814; http://www.councilscienceeditors.org) or other standard sources.

The table below lists standard accepted abbreviations for typical cleft-type classifications and study groups. Other abbreviations may be proposed for classifications and groups not listed.

4.6.3 Tables

Tables should be numbered consecutively using Arabic numerals. Each table should have an appropriate title and explanation at its head. Abbreviations used in a table must be explained in a footnote below the table. Submit tables as separate files, with one table per file, in either .doc (text) or .xls (spreadsheet) format.

4.6.4 Figures

All figures and illustrations must be original photographs or artwork. For figures or illustrations reprinted from published work, the author must obtain written permission from the copyright holder and disclose that upon submission. Submit figures as separate files.

Illustrations, pictures and graphs should be supplied in the highest quality and in an electronic format that helps us to publish your article in the best way possible. Figures submitted at lower than the required resolutions stated above will be allowed for review purposes. However, the publication process for accepted manuscripts will be delayed until acceptable images have been submitted. Please follow the guidelines below to enable us to prepare your artwork for the printed issue as well as the online version.

- **Format:** TIFF, JPEG: Common format for pictures (containing no text or graphs).
  EPS: Preferred format for graphs and line art (retains quality when enlarging/zooming in).
- **Placement:** Figures/charts and tables should be submitted separately. Please add a placeholder note in the running text (i.e., “[insert Figure 1.]"). A single figure may include multiple images (a, b, c, etc.) but all must appear on the same page. Figures should be numbered consecutively in the order in which they appear in the manuscript, using Arabic numerals (e.g., Figure 1, Figure 2, etc). Figure legends must be included on a separate page following the body of the manuscript. The legends should explain each figure in detail.
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- **Color:** Please note that images supplied in color will be published in color online and black and white in print (unless otherwise arranged). Therefore, it is important that you supply images that are comprehensible in black and white as well (i.e., by using color with a distinctive pattern or dotted lines). The captions should reflect this by **not** using words indicating color. For specifically requested color reproduction in print, you will receive information regarding the costs from SAGE after receipt of your accepted article. The first color image is $800, and it is $200 for any additional color images within the same contribution.
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Image Integrity

Figures should be minimally processed and should reflect the integrity of the original data in the image. Adjustments to images in brightness, contrast, or color balance should be applied equally to the entire image, provided they do not distort any data in the figure, including the background. Selective adjustments and touch-up tools used on portions of a figure are not appropriate. Images should not be layered or combined into a single image unless it is stated that the figure is a product of time-averaged data. All adjustments to image date should be clearly disclosed in the figure legend. Images may be additionally screened to confirm faithfulness to the original data. Authors should be able to supply raw image data upon request. Authors should also list tools and software used to collect image data and should document settings and manipulations in the Methods section.

Visual Abstracts

A graphical abstract is meant to be a clear, quick, and concise pictorial representation of research that has been published in the journal. It is meant to support the written abstract that accompanies all papers submitted for review to the journal. All figures published in the journal, including graphical abstracts, should be of the highest quality and should highlight paper findings. Please note visual abstracts are optional, but if you wish to submit a visual abstract with your paper, please follow the below guidelines:

- The graphic should be labelled as “graphical abstract” or similar, so that it is clear the file is not an article figure file (e.g., it should not be labelled “Fig1”, “Fig2” etc.)
- The aspect ratio for the graphic should be 16:9 (the recommended size ratio would be 600px X 338px)
- The figure file type should be the same as for other article figures. Graphical abstracts, as with all figures in the journal, are only accepted in the following formats: JPG, TIF, or EPS. The journal does not accept Word or PowerPoint figure files.
- A caption should be provided with the graphic. The caption should read: “This is a graphical representation of the abstract”
- Do not use images subject to copyright clearance for graphical abstracts. If at all, graphical abstracts should feature aspects of the original figures created for the paper it is supporting.
- The final visual abstract image should be sent with accepted article.
- Simplicity is the key to conveying information visually. Terms and abbreviations should match overall journal usage and style.

4.6.5 Supplemental Material

This journal is able to host additional materials online (e.g., datasets, podcasts, videos, images, etc.) alongside the full text of the article. For more information, please refer to our guidelines on submitting supplementary files.

Supplemental figures, tables, data files, and text
These types of supplemental files should be named as Supplemental followed by the number in the sequence (e.g., Supplemental Figure 1; Supplemental Table 1) and referred to in the body of the manuscript text.

Video

Video clips that contribute significantly to the manuscript may be submitted in either avi, mov, or mpeg formats. Videos should be submitted at the desired reproduction size and length but should not exceed 10MB in size. If submitting avi files, the files must be compressed. Authors are solely responsible for all editing of video clips.

As there are restrictions to the video file size, we recommend compressing the file and uploading it to the CPCJ SAGE Track platform. The manuscript review system ScholarOne has a file size limit of 350mb for video files. If the video you wish to submit for review is larger than this, please follow these instructions on compressing the video file to fit within this limitation.

Please note that if your submission is accepted, you will be asked to provide the full-size file for publication. This can be provided to production via DropBox or Google Drive.

Each video file must be accompanied by a still image from the video that conforms to the figure resolution and size requirements outlined above for figures. This image will be published in the print version of the journal in place of the video. Please indicate in the figure legend that the still image has an associated video file. Both the print-version figure and the video must share the same file name (e.g., Figure1.jpg and Figure1.mov). A "List of Video Legends" should be prepared on a separate page at the end of the manuscript article file.

Video submissions are strongly encouraged, particularly for articles dealing with surgical techniques.

For more information about the format requirements for videos, please review our Author Gateway. For detailed information pertaining to copyright and permissions requirements, view the Video Permission and Fair Use Quick Guide.

For videos with identifiable subjects, subjects will need to sign the Audio- Visual Likeness Release Form. **It is the author’s responsibility to submit signed release forms, if necessary, for each video.** If patient(s) are identifiable in the video, authors must confirm a Patient Permission form has been completed and signed by each patient.

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Best-practice guidelines for preparing videos are be found at the following link: https://jamanetwork.com/journals/jamapediatrics/pages/instructions-for-authors.

Audio
Audio clips that contribute significantly to the manuscript may be submitted in .au, .ram, .wav, or .mp3 formats. Audio files should not exceed 6 MB in size. Authors are solely responsible for all editing of audio clips. Audio clips should be cited in the manuscript as Audio 1, Audio 2, etc. A "List of Audio Legends" should be submitted on a separate page at the end of the manuscript article file.

4.7 English language editing services

Authors seeking assistance with English language editing, translation, or figure and manuscript formatting to fit the journal's specifications should consider using SAGE Language Services. Visit SAGE Language Services on our Journal Author Gateway for further information.

4.8 ORCID

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The collection of ORCID IDs from corresponding authors is now part of the submission process of this journal. If you already have an ORCID ID you will be asked to associate that to your submission during the online submission process. We also strongly encourage all co-authors to link their ORCID ID to their accounts in our online peer review platforms. It takes seconds to do: click the link when prompted, sign into your ORCID account and our systems are automatically updated. Your ORCID ID will become part of your accepted publication's metadata, making your work attributable to you and only you. Your ORCID ID is published with your article so that fellow researchers reading your work can link to your ORCID profile and from there link to your other publications.

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For further information including guidance on fair dealing for criticism and review, please see the Copyright and Permissions page on the SAGE Author Gateway.

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5. On acceptance and publication

5.1 SAGE Production

Your SAGE Production Editor will keep you informed as to your article's progress throughout the production process. Proofs will be made available to the corresponding author via our editing portal SAGE Edit or by email, and corrections should be made directly or notified to us promptly. Authors are reminded to check their proofs carefully to confirm that all author information, including names, affiliations, sequence and contact details are correct, and that Funding and Conflict of Interest statements, if any, are accurate.

5.2 Online First publication

Online First allows final articles (completed and approved articles awaiting assignment to a future issue) to be published online prior to their inclusion in a journal issue, which significantly reduces the lead time between submission and publication. Visit the SAGE Journals help page for more details, including how to cite Online First articles.

5.3 Access to your published article

SAGE provides authors with online access to their final article.

5.4 Promoting your article

Publication is not the end of the process! You can help disseminate your paper and ensure it is as widely read and cited as possible. The SAGE Author Gateway has numerous resources to help you promote your work. Visit the Promote Your Article page on the Gateway for tips and advice.

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6. Further information

Any correspondence, queries or additional requests for information on the manuscript submission process should be sent to the *The Cleft Palate Craniofacial Journal (CPCJ)* editorial office as follows:

Editor: Jamie Perry, PhD
Editorial Office: The Cleft Palate Craniofacial Journal
Email: perryja@ecu.edu

If you have any questions about publishing with SAGE, please visit the SAGE Journal Solutions Portal

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7. Appealing the publication decision

Editors have very broad discretion in determining whether an article is an appropriate fit for their journal. Many manuscripts are declined with a very general statement of the rejection decision. These decisions are not eligible for formal appeal unless the author believes the decision to reject the manuscript was based on an error in the review of the article, in which case the author may appeal the decision by providing the Editor with a detailed written description of the error they believe occurred.

If an author believes the decision regarding their manuscript was affected by a publication ethics breach, the author may contact the publisher with a detailed written description of their concern, and information supporting the concern, at publication_ethics@sagepub.com

Appendix F

MDT Survey Questions

1. In your opinion, what makes the decision about surgery easy or difficult for parents? *Please provide as much detail as possible.*

2. In your opinion, what are the most important sources of information available to parents to aid decision making? *Please provide examples if possible.*

3. In your experience, what other processes do parents go through to reach a decision about corrective surgery?

4. What information is it important for you / your speciality to convey to help parents reach a decision? *Please provide as much detail as possible.*

5. What do you feel is currently being done well in the service to help parents reach a decision regarding corrective surgery? *Please provide examples if possible.*

6. What do you think could be done within the service to improve the decision-making process for parents?

7. The concept of ‘shared decision-making’ is becoming increasingly important in the NHS. Shared decision making is a joint process in which a healthcare professional works together with a person to reach a decision about care. It involves choosing tests and treatments based both on evidence and on the person's individual preferences, beliefs and values. It makes sure the person understands the risks, benefits and possible consequences of different options through discussion and information sharing.

   What do you as an individual clinician do to promote shared decision making?

8. What do you as an MDT do to promote shared decision making?

9. Is there anything you think could be done better to promote shared decision making?

10. Patient decision aids are a means of helping people make informed choices about healthcare. A decision aid will usually provide written or visual information which can include the following:
    a. The treatment or care options recommended in NICE guidance.
    b. The aims of treatment or care and how likely the person is to benefit.
    c. Possible adverse effects from the treatment or care options and the likelihood of experiencing them.
    d. Other issues likely to be important to the person facing the decision (such as additional monitoring requirements and duration of treatment).

    Do you feel having a decision aid would be of help to parents?

11. Why do you think a decision aid would / would not be helpful?

12. Is there anything else you think it would be helpful for us to know?

Appendix G

Parent Survey Questions

1. What steps did you go through to reach a decision regarding surgery for your child? Please take time to reflect and provide as much detail as possible.

2. What guided your thinking about surgery as an option? Please tick all of the following options which apply:
   i. Appearance.
   ii. Risk of increased intracranial pressure.
   iii. Concerns about development
   iv. Scarring.
   v. Risks of surgery.
   vi. Other – please specify.

3. Which of these felt particularly important to you?

4. How did you feel about the idea of deciding to pursue surgery for your child? What emotions did you feel while making this decision?

5. Who else did you seek an opinion or advice from prior to making your decision? How important was this to your decision?

6. Did you find making the decision particularly easy or difficult? What made it easy or difficult?

7. At what point did you feel as though you had made your decision to pursue surgery for your child?

8. What did the service do well in helping you to reach a decision?

9. The concept of 'shared decision-making' is becoming increasingly important in the NHS. Shared decision making is a joint process in which a healthcare professional works together with a person to reach a decision about care. It involves choosing tests and treatments based both on evidence and on the person's individual preferences, beliefs, and values. It makes sure the person understands the risks, benefits, and possible consequences of different options through discussion and information sharing.

   Do you feel like the craniofacial team promoted shared decision making?

10. If yes, what do you think the craniofacial team did to promote shared decision making?

11. Is there anything you think could be done better to promote shared decision making?

12. "Patient decision aids are a means of helping people make informed choices about healthcare. A decision aid will usually provide written or visual information which can include the following:
    - The treatment or care options recommended in NHS guidance.
    - The aims of treatment or care and how likely the person is to benefit.

  - Possible adverse effects from the treatment or care options and the likelihood of experiencing them.
  - Other issues likely to be important to the person facing the decision (such as additional monitoring requirements and duration of treatment).

Would a decision aid have been helpful during the decision-making process?
Why do you think a decision aid would / would not have been helpful?

13. Is there anything else you think it would be helpful for us to know about how you found the decision-making process?

Appendix H

Bracketing Interview Questions and Excerpts (please note, some of the interview data has been removed as it is of a personal nature)

Bracketing interview 1 – Before data collection

1. What drew you to this topic?

"...I have always been interested in medicine and hospitals, maybe this comes from loving things like Casualty and Grey's Anatomy growing up. I was always fascinated by things like 24 Hours in A&E and wondered how on earth people were coping with such life changing injuries. During my GCSE's I actually thought I wanted to be a Doctor because I thought I would thrive being in a hospital setting…. Obviously with Mum, I was also in and out of hospitals growing up so was exposed to and helped by loads of medical professionals. To me, hospitals are a place of physical and psychological pain but also a place of care and incredible support of people at what might be the most difficult time in their lives. I think this is something I always wanted to be a part of. I guess, to give back to families like our own..."

2. What assumptions are you making, before analysing the data?

"….I am definitely assuming that this will be an unbelievably difficult decision for parents to make. I am indecisive by nature so imagine parents will go backwards and forwards, as I would in making any decision, maybe making a pros and cons list or talking through the details with a loved one…."

3. What steps would you go through to make a decision?
4. What steps would you go through to reach a decision regarding surgery for your child?

"… It is obviously important to acknowledge that I don't have children so can only imagine how someone would react and feel during such a decision. I imagine I would feel hugely protective over my child, as well as a huge responsibility to make the right decision for them and their future, not that you will ever necessarily know if the decision you make for them is the 'right' decision…"

5. What would guide your thinking about surgery as an option?

"…I don't like to admit it, and perhaps feel as though I shouldn't, but I think appearance would matter a lot to me. Through placements, I have seen how cruel children can be to those who are different in any way. Particularly in my CAMHS placement in a neurodevelopmental conditions team, every child I worked with had experienced bullying because they were seen as 'different'. I saw first hand the impact that this then had on their confidence, self-esteem and mental health. I wouldn't want my child to grow up the victim of bullying…"

6. How would you feel about the idea of deciding to pursue pursuing surgery for your child?
7. Who else would you seek an opinion or advice from prior to making a decision?
8. Would you find the decision particularly easy or difficult?

"…I would think that the decision is easier for parents who are very risk averse and don't want to take the risk of not undergoing surgery or who care significantly about physical appearance. I would assume that for parents who are leaning towards not having the surgery, the decision is harder…. I would think that the decision may be harder for people from different countries. I imagine trusting doctors or surgeons from a country other than your home country would be challenging. I would imagine that the decision would be easier for those who have had previous positive experiences of healthcare, and harder for people with

more negative experiences of accessing healthcare, or maybe surgery that had had unexpected complications in the past…"

Bracketing interview 2 – After data collection

1. What feelings arose for you during the data analysis period?

"… The first time I read through the parent data, I felt really quite moved by the responses. I could see how emotionally challenging these parents had found the process, but they were able to reflect on how the team had helped them through this. The things they had to say about the team were absolutely incredible and it made me really glad that parents had been able to share their thanks on this platform, and that this would be fed back to the MDT…"

2. Did any of your assumptions change throughout the data analysis period?
3. How has working with the Craniofacial Psychologists on placement impacted your views on parents proceeding (or not proceeding) with surgery?

"… For me, it has confirmed in my mind that surgery is something I think I would proceed with, should I have a child with craniosynostosis. I know that all of the parents who completed the survey were ones who opted for surgery themselves and I can't help but thinking this will bias results. Not only would I opt for surgery, but also all of the parents who completed the survey would too…"

Appendix I

MDT Codes, Sub-themes and Themes

Theme	Sub-theme	Code	Files	References
1. Background factors		Need for flexibility	4	4
		Child as an individual	3	4
		Individual differences in parents	2	3
		Tailored amount of information given	5	7
	1.1 Personal Factors	Attitudes towards risk	2	4
		Personal values around appearance	5	8
		Previous parental experiences	2	2
	1.2 Demographic factors	Race/ethnicity impact	1	1
	1.3 Creating the right environment for decision making	Time pressure of NHS appointments	6	8
		Time pressure to make a decision	2	2
		Allowing space for feeling	2	2
		Providing reassurance	2	4
		Discussion (no right or wrong)	6	12
		Multiple appointments if needed	7	14
		Reflection opportunities	4	11
		Taking time to make the decision	4	8
		Clinical Psychologist’s role	7	13
2. Attitude towards the behaviour	2.1 Instrumental attitude	Clear information provided	4	7
		Consistency	3	3
		Evidence based information	2	3
		Importance of detail	2	3
		Other centre’s options	3	5
		Information provided in clinics	7	16
		Written information to help remember details	6	7
		Visual information	6	8

		Positive response to decision aid	10	11
		Peer support	9	16
		Social media	7	8
		Inaccurate information	4	5
		Charity support	5	6
		Bullying fear	2	2
		Developmental considerations	3	7
		Future impacts considered	5	6
		Aesthetic reasons for surgery	6	8
		Raised intracranial pressure	5	6
		Information about follow up if don’t opt for surgery	5	5
		Ensuring understanding	8	15
		Nursing role	8	13
		Logical approach to decision making	2	2
		Weighing up pros and cons	6	13
	2.2 Experiential attitude	Making the ‘right’ decision	1	1
		Emotional toll of decision	5	10
		Easier decision	9	13
		Harder decision	7	10
3. Subjective norm	3.1 Normative beliefs of others	Discussion between parents of child	5	6
		Child’s voice (or lack of)	2	4
		MDT opinion	4	10
		Discussion with loved ones	7	9
	3.2 Motivation to comply	Societal norms	1	1
4. Perceived control	4.1 Promoting parent’s control	Trust between the MDT and the family	2	2
		Alongside parents	1	1
		Parent’s voice	7	14

Appendix J

MDT Quotes

Theme	Sub-theme	Quotes
1. Background factors	1.1 Personal Factors	“I think it depends on the level of uncertainty a parent is comfortable with and their perceptions around appearance also.” (MDT10) “I think it depends often on perceptions of risk and chance, and individual differences in how people assess these factors and what is an acceptable level of risk for one set of parents, which may totally differ to another.” (MDT5) “Weighing up their own personal priorities relating to appearance.” (MDT1) “If a parent has their own experience or is aware of other's experiences of negative reactions around craniofacial difference, they may feel that they want to prevent this from happening to their child by opting for surgery.” (MDT10) “For other parents who are considering the aesthetic reasons for having surgery, the decision is also easy as they feel strongly, due to their own experiences and beliefs that this will make their child's life better.” (MDT5)
	1.2 Demographic factors	“I do think there is some intersectionality with race/ethnicity in terms of making the decision. For example, one father was particularly worried about his son, who was black, growing up with a scar on his head and how this may affect his future job prospects.” (MDT3)
	1.3 Creating the right environment for decision making	“The time constraints of NHS appointments are difficult.” (MDT1)

“In the past, I used to see some families separately with the clinical psychologist straight after their initial appointment with the Consultant and go over information and options. This hasn't happened since Covid, due to time constraints and various other factors. I do think it was helpful though for some families.” (MDT2)

“Clinic is quite a stressful environment for parents, with lots of information given, and lots of professionals in the room. I wonder whether there is any way parents could have an opportunity to discuss surgery and their questions without all of us there? Side room discussion? Not sure how feasible this is.” (MDT9)

“Give them plenty of time to think.” (MDT7)

“Speaking to parents and listening to their fears and anxieties and trying to alleviate these.” (MDT2)

“Listen to their concerns and validate, normalise and reassure with the information we do have available” (MDT3)

“Consider what is important to families, their values and beliefs and make it clear that the decision is theirs. Offer non-judgement about which option is taken.” (MDT5)

“Several families have expressed gratitude for having this discussion element available during the pre-operative process.” (MDT3)

“Provide all the information in an unbiased way and ensure the parents understand it is not an instruction, or even a recommendation, but a choice for them that we will support.” (MDT6)

“Offer for them to be contacted by individual members of the MDT separately.” (MDT7)

2. Attitude towards the behaviour	2.1 Instrumental attitude	“Information should be of high quality and very specific to that patient, where possible.” (MDT8) “Parents are given clear information about the diagnosis, clear information about the surgery.” (MDT1) “Keep close contact with the rest of the team to make sure we are all approaching from the same perspective and sending consistent messages.” (MDT3) “Providing evidence based information required to aid decision making rather than leaving families to search out information, possibly from biased or inaccurate sources.” (MDT5) “Access to overview information about how all of the centres provide care and surgery to allow parents to make a choice between centres e.g. if exploring endoscopic suturectomy and helmeting.” (MDT9) “[In clinic, we give] information about the underlying diagnosis and relative risk of ICP, eye difficulties, developmental difficulties etc., genetic information to aid a better understanding of their child's decision, understanding about their child's current development, information about the invasiveness of the surgery and the relative risks, information about what the surgery will actually rectify.” (MDT1) “A written summary of information could help provide a balanced view that families can refer to at a later date and share with others to help them make the decision.” (MDT3) “I do think the provision of additional written information about the surgery could help parents reflect on these important factors after an appointment.” (MDT1) “Pre and post op photos very useful for placing into perspective.” (MDT4)

	“Support from other families who have gone through surgery (or not) is likely to enable decision making.” (MDT8)
	“Other families turn to facebook groups, or even other families who are staying in the same accommodation (Ronald McDonald House) to hear the perspective of others who have been through the process already.” (MDT3)
	“I am aware patients use social media, which I think is important as it provides personal experience from others who have gone through the same process, but I do worry about how unregulated this is, and that parents may be receiving information that is not relevant to their child, or accurate.” (MDT9)
	“I'm sure most people will search google images which can produce some extreme cases, and some inaccurate information of how things are done in other countries.” (MDT3)
	“We will continue to support them whether they choose to have surgery or not.” (MDT9)
	“Supporting them in making the decision using a framework to structure their thinking.” (MDT5)
	“I think many parents get all the information they can and weigh things up, looking at the balance of risks and probabilities. However, other parents make much quicker decisions based on a strong sense of what is the best thing for their child.” (MDT10)
2.2 Experiential attitude	“As the children are young and do not yet have their own voice, parents feel the pressure to make the right decision for their future, which is a big responsibility.” (MDT3)
	“I often see how this decision weighs on parents' minds…In these cases where the risk of raised intracranial pressure is low and the primary reason for surgery is aesthetic, I think parents sometimes feel guilty for seemingly wishing to change the look of their child and exposing them to the (relatively low) risks of surgery when it is not absolutely necessary.” (MDT3)

		“I think severity of deformity makes it easier - parents can imagine the child would have a harder life with significant facial deformity.” (MDT6) “I think that once the risk of high pressure is mentioned, this makes the decision 'easier' for the parents as parents do not want to run the risk of their child developing high pressure.” (MDT2) “Parents with quite black and white thinking, or who think about the process purely in terms of logic and statistics, tend to have less trouble making the decision.” (MDT3)
3. Subjective norm	3.1 Normative beliefs of others	“They will often discuss between partners and wider family, before coming to a decision….I therefore wonder if this can be harder for more isolated single parents.” (MDT3) “Being able to discuss the decision with each other (both parents, or parent and their partner or family).” (MDT9) “I think the agonising part of surgical decision making for parents is trying to make a decision that they would like to have their child's voice and views on, when their child is too young to participate themselves.” (MDT1) “I think the most difficult decisions are in mild cases where the MDT do not want to strongly recommend to proceed with surgery or not.” (MDT3) “Even though some parents would like us to make the decision for them, we resist to encourage them to take some ownership of it, with our support.” (MDT6)

	3.2 Motivation to comply	“They almost always want what's best for their child growing up so they are not bullied or feel visibly different, but then can simultaneously love their child for the way they look. This means the primary function of the surgery is to allow their child to conform with societal norms which can be an uncomfortable position to take as a parent. I think thoughts of ""why should I force my child to go through a serious surgical procedure just so they'll fit in?"" are often contrasted with thoughts of ""who am I to deny my child their right to a 'normal' childhood?"". Of course, an added complication is that the child will then grow up with a scar on their head, so there is a potential option for visible difference either way, which is especially difficult when the visible difference in head shape is mild.” (MDT3)
4. Perceived control	4.1 Promoting parent’s control	“Establishing a strong rapport and clinical relationship with the patient's family is key.” (MDT8) “Trust in the professionals caring for their child.” (MDT9) “Speak frequently to parents whilst they are on the ward about the care their child is receiving. Liaise with ward staff and be the family's advocate.” (MDT2) “Eliciting parent's views to a greater extent in this setting also rather than more focus on didactic information giving…Taking time to listen to the parent's views and the values and beliefs underpinning this, so that I can understand their position. If I understand their position, I am in a better place to aid them in deciding. I may also advocate for a family, representing their views to the wider MDT” (MDT10)

Appendix K

Parent Codes, Sub-themes and Themes

Theme	Sub-theme	Code	Files	References
1. Background factors		Not just a number	1	2
	1.1 Personal factors	Bury head in the sand	1	1
		Risk tolerance	1	1
		Previous surgeries	2	2
	1.2 Demographic factors	Gender differences	1	1
2. Attitude towards performing the behaviour	2.1 Instrumental attitude	Aesthetic concerns	15	30
		Developmental concerns	15	20
		Intracranial pressure worries	18	39
		Risks of surgery or no surgery	7	11
		Hypervigilance as they grow up	3	4
		Pictures	3	3
		Clear understanding of all the necessary information	15	28
		Tests pre-surgery	2	4
		Answering questions from parent	12	19
		Facebook	6	8
		Own research	8	11
		Speaking to other parents	8	13
		Watching documentary	1	1
		Positive response to decision aid	5	5
		No need for decision aid	5	5
		Changes to condition	3	3
		Only option	10	11
	2.2 Experiential attitude	Emotions caused by the decision	17	31
		Possibility of losing your child	5	7
		Making the ‘right’ decision	6	6
		Cancellation possibility	1	1

		Covid impacts	1	2
		Waiting for surgery	1	3
		Currently health child	2	3
		Prepared before meeting MDT	5	7
	2.3 Inner conflict between instrumental and experiential attitude	Conflict between emotion and cognition	4	5
3. Subjective norm	3.1 Clashes of normative beliefs	Bullying fears	8	11
		Discussion between parents	6	9
	3.2 Motivation to comply	Family and friends' opinions	9	12
4. Perceived control		Decision point	16	18
		No rush to make a decision	5	6
		Parent's decision to make	6	6
		Patience of the MDT	6	6
5. "Without every single one of them, we would have been lost"		Caring MDT	4	6
		Compliments for the MDT	7	12
		Exceptional service provided by the MDT	6	11
		Expertise provided by the MDT	6	11
		Advantage of having a multi-disciplinary team	2	3
		Reputation of the John Radcliffe Hospital	3	4
		Trust in the MDT	4	4

Appendix L

Parent Quotes

Theme	Sub-theme	Quotes
1. Background factors	1.1 Personal factors	“In theory, our son might never experience issues caused by his head shape, but we considered this to be a gamble we weren't prepared to make.” (P3) “Its a difficult decision understanding the tender age and with the previous few surgeries experiences made it more difficult decision.” (P2) “Having been through one major surgery almost made the decision for her second a little easier.” (P10) “Bullying because I was bullied my entire childhood for something I had no control over and I didn’t want him to go through that as well.” (P12)
	1.2 Demographic factors	“Although fundamentally bullying was in the back of my mind being female and as she would get older she would look different to her piers.” (P1)
2. Attitude towards performing the behaviour	2.1 Instrumental attitude	“Appearance we felt was quite important to us also as even though we wouldn’t mind either way how he looked or could possibly look we just didn’t want this to be a problem by others for him as he grows.” (P4) “Primarily it was the balance between the risks of surgery against the potential impacts on development, speech and vision if left untreated.” (P3)

“Not having the risk of increased pressure and the development issues. We both felt these would be major factors in having surgery so we could stop him developing pressure and stopping any development issues later down the line if he didn’t have surgery.” (P7)

“My biggest issue was if she didn’t have the surgery in the future this may affect the pressure in her brain and give her issues.” (P1)

“Would we be worried every time he had a headache or had an episode of staring if we did not operate.” (P5)

“With the risk of intracranial pressure, we didn’t want to take any risks to prolong this that could potentially be fatal.” (P10)

“When we met the Oxford team and they explained about the risk of inter cranial pressure that was the key point for us in agreeing to go forward with surgery as we didn't want to risk that.” (P16)

“It was definitely intracranial pressure. The thought of not knowing whether she would or would not develop learning difficulties or more as a result of increased intracranial pressure.” (P17)

“He had an episode of unusual movements and although it was felt not to be related, we were concerned and thought we would have worry of pressure in the back of our minds throughout his childhood.” (P5)

“We just knew that we would rather him have the surgery than live life with a what if something happened.” (P6)

“We got shown a lot of pictures of children before & after surgery, but these pictures were while the children still very young. No pictures of children older than 5 or 6, no pictures of children who chose not to have surgery at an older age & nothing to show teenagers or adults which is what we wanted to see, not how this will affect our son now, but how it will affect him in 10-20 years time.” (P15)

“Taking every stupid questions of us and giving the detailed answer with loads of patience and making sure we are absolutely clear and understand the process….Giving us the insight of the future possible complications and challenges and with multiple tests to support the decision.” (P2)

“All risks were explained of both surgical correction and not operating.” (P5)

“Offered all eventualities and allowed us to make our own decision with all the information they could give us.” (P9)

“[Dr] handled our stress very well, answer all the questions we had according to his experience, the most important thing is he did not give us any hopes , he came up to us very realistic way - that he will do his job as good as he can but not everything is up to him . His experience and knowledge encouraged us to go for the surgery.” (P11)

“They offered as much information available in regards to the benefits & risks. They were happy to answer any questions with honesty & discuss any concerns whether it be during appointments or via email.” (P15)

“We did alot of research and joined the Facebook group cranio ribbons to see other families experiences, to see before and afters and just to talk to other parents that went through the same surgery, that really helped a great deal in knowing we made the right decision.” (P8)

“Using a support group (cranio ribbons) to speak to other parents who have or not chosen to have surgery, using their experiences to come to a decision.” (P15)

“We looked at some of the parents support facebook groups (e.g. cranio ribbons) but largely felt that these were quite anxious places that weren't helpful for us. Having said that I did post once on there to ask for practiccal advice about people's hospital stays and one mum did message me sharing lots of information about her experience of hospital and the operation including sharing photos.” (P16)

	“Within the three months before even meeting the surgeon as I had prepared and researched the condition made a point of doing all that I could to understand.” (P1) “Own research into the condition (using the internet), including watching videos of the surgery and looking at before/after pictures. Researched [surgeon’s] history, including news articles. Reviewed the Care Quality Commission reports and Integrated Performance Reports (all available online). Once we were satisfied that OUH was a competent hospital, we considered whether the operation was necessary.” (P3) “I think having all the information easily accessible would help any decision making process.” (P9) “For us it wasn’t really a debate, we both agreed we wanted the surgery for him to improve his head for practical reasons and cosmetic.” (P9) “After doing research, it didn’t feel there was any other option than for him to have surgery.” (P15)
2.2 Experiential attitude	“With 5 months to prepare and wait for the date I have never found life so difficult. No matter how much I tried to rationalise myself- the risks of surgery and taking a babies skull of I could help but prepare for her to die. Overwhelmed sadness a child so small had to go through something so horrendous. In those 5 months my life came to a standstill. And nothing could help other than be the other side of the surgery to know she is alive.” (P5) “The emotions that we experienced were stress, anger, fear and anxiety.” (P3) “A lot of emotions! Particularly worried, scared, upset.” (P4) “The obvious emotion we shared was apprehension and anxiety, also slight guilt.” (P6)

	“We both felt emotional as at the time we had come to the initial decision he was only 4 months old so he was still only small. However we both knew it needed to be done even if the whole processes would be upsetting and emotionally draining we would still go ahead with it for the benefit of our son.” (P7)
	“The emotional side to it all was a huge rollercoaster of ups and downs, sometimes it could make you feel sick where the worry and anxiety got too much.” (P8)
	“I felt a lot of pressure to make the correct decision on behalf of my child. A lot of fear & anxiety around him having to have major surgery at such a young age.” (P15)
	“I also had feelings of guilt as somehow I thought it was my fault she was born with this.” (P17)
	“Then came the operation maybe cancelled due to beds, and two specialities were fighting over the one bed- in that hour the decision being made I remember almost floating in the room as it was so horrendous preparing for the day I couldn’t comprehend that a bed situation was going to possibly delay for the surgery.” (P1)
2.3 Inner conflict between instrumental and experiential attitude	“It was still a very difficult decision on the basis that we were exposing our completely healthy son to a procedure that could result in paralysis, blindness, disability and death. Despite the rational approach we had taken to our decision, there is still a significant emotional barrier to overcome when signing the forms….The rational element of the decision making process was relatively easy, overcoming the emotional barriers was difficult.” (P3)
	“We both felt emotional as at the time we had come to the initial decision he was only 4 months old so he was still only small. However we both knew it needed to be done even if the whole processes would be upsetting and emotionally draining we would still go ahead with it for the benefit of our son.” (P7)

3. Subjective norm	3.1 Clashes of normative beliefs	"As part of our parenting approach we also want our children to understand that physical looks don't define who you are and we want them both to have a strong sense of self worth. There was a small part of us that obviously was aware that the wider world (e.g. school peers potentially bullying in future) wouldn't all share this perspective and so that element of the aesthetic issue did feel more important." (P16) "We also thought a lot about how his head shape may effect him not just physically but emotionally when at school if left the way it was." (P8) "We felt that sometimes other children can be unkind and you could tell that our son had a flat side to his head and his eye was not right." (P14) "It was a tough decision but we spoke about it together as a couple and as a family in such great detail." (P5) "We spoke to each other (myself and my husband - his father). We both outright separately decided surgery would be best option for us but wanted to make sure both of us agreed." (P7)
	3.2 Motivation to comply	"We spoke to both our families but it wasn't changing out decision we just needed to talk it over further with other people." (P7) "We are a very close family. They were all in agreement that both surgery's were important, but would support us in whatever decision we made." (P10) "Family members and friends. Decision had already been made but they reassured me and said that it was in the best interest of our son." (P14)

	“Another thing to note was that whilst we felt certain about surgery wider family members or friends questioned why we needed to put her through this. This was often coming from a place of trying to save us from going through a difficult experience but it did feel at times like people were questioning our judgement as parents which did sometimes make us second guess our approach…We didn't seek opinions from family and friends but they were sometimes offered unsolicited. These didn't affect our decision making.” (P16)
4. Perceived control	“When we signed the form. Up to that point we still felt the decision had not crystallised.” (P3) “We felt we made the decision after speaking to [surgeon] from Oxford cranio facial team in detail about the surgery and not having the surgery. Once we went away and took everything in that we was told and spoke about once we had discussed it together it was clear what our decision would be.” (P4) “We decided immediately we wanted the surgery.” (P9) “The decision was made as soon as we were explained the risks if we chose not to have the surgery.” (P10) “I questioned right up to the last hour if it was the right thing to do for him.” (P15) “After his first appointment we made the decision.” (P7) “The same day we were advice that surgery was on the cards. Decision was made during consultation.” (P17) “They explained the different options we could take and what they recommended but at the end of the day the decision was ours.” (P7) “They gave us all the facts and didn't pressure us into making a quick decision.” (P17)

	“Offered all eventualities and allowed us to make our own decision with all the information they could give us.” (P9) “They provided all the facts & shared their opinions on the best options available. Giving us as parents, the final decision.” (P15) “They were always very clear we had a choice about surgery.” (P16)
5. “Without every single one of them, we would have been lost”	“The JR’s specialist nurse is one of the most amazing women I have ever met. She was exceptionally patient and reassuring and it was wonderful every time we went to the JR she would make a point of touching base. [Child] wasn’t ‘just another patient’ [nurse] cared and made us feel as though she was with us on the journey. [Nurse] herself is the one person who gave so much support to us as a family- I will never forget this.” (P1) “Without every single one of them we would have been lost….I think you all should know how amazing you all have been.” (P10) “They handled our stress very well , were patient, answers all the questions with all their experience and knowledge. We have met right people in the right place . We can’t thank them enough !!!” (P11) “The Cranio service works like clock work and is not like any other service Iv ever experienced in the NHS….All staff very confident and experienced in their fields, never did I feel as though they didn’t know any answers to my questions….Speach and language therapist [name] is also exceptional and it makes me smile watching her interact with [child] as she is clearly expert in her field.” (P1) “I knew he was going to be in the best hands.” (P8) “Team was very approachable and knowledgeable.” (P17)

“Providing access to experts from a number of different disciplines.” (P3)

“You have to trust complete strangers doing surgery on your child and scared if complications happen.” (P18)

Appendix M

Author Guidelines for Body Image

Types of Papers

The journal publishes

1. Full-length articles of the following types: Original research articles (studies that do not fit one of the other types listed below) **Systematic reviews / meta-analyses** (please follow PRISMA checklist: http://www.prisma-statement.org/ **Methodological / protocol articles** (articles that explicate an innovative research study design in which data are currently being collected) **Unexpected / null results articles** (articles grounded in extant theory that have a sound methodological design and adequate statistical power and are analyzed appropriately, but primary hypotheses were not supported) **Scale development / adaptation articles** (multi-study/sample articles that investigate the psychometric properties of a newly developed or existing scale relevant to body image; scale translations and applications to different samples are welcome) **Replication studies** (consistent with Open Science initiatives, we encourage articles that replicate--or fail to replicate--existing body image research) **Theoretical review articles** (typically invited; however, if you have an idea, propose it to the Editor-in-Chief)

Please choose the article type that is the best fit for your article (we realize that some articles may fit into more than one type).

While full-length articles have no explicit limits in terms of numbers of words, tables/figures, and references, an article's length must be justified by its empirical strength and the significance of its contribution to the literature.

2. Shorter communications of the following types: Brief research reports (articles with a more defined and/or limited focus than original research articles) **Ideas worth researching** (articles that propose a novel idea for advancing research on body image) **Methodological innovations** (articles that discuss the application of a novel statistical approach to the study of body image)

Guidelines for short communications are <= 3000 words from Introduction through Discussion and <= 30 references. There are no limits on tables and figures

3. Themed special issues Theoretical special issue (a collection of review articles from experts in the body image field that focus on a relevant body image topic) **Empirical special issue** (a collection of empirical articles that offer novel insights into a relevant body image topic) **Data set special issue** (a collection of empirical articles that emerge from the same, large data set; each article within the issue must be incremental and overlapping data between articles must be minimal)

We especially encourage special issues that bridge body image theory and research with other disciplines and social science constructs.
Please contact Editor-in-Chief to propose your idea for a special issue.

If you are proposing a theoretical or empirical special issue and it is accepted, you will be the Guest Editor(s) and work with the Editor-in-Chief (or an Associate Editor) and our Special Issue Journal Manager to develop and prepare your special issue.

If you are proposing a data set special issue, then Guest Editors will be appointed that manage your issue and they will work with the Editor-in-Chief (or an Associate Editor) and our Special Issue Journal Manager.

For each paper type, we would like authors to know that we are impartial regarding the source of citations and we recommend against excessive string citations.

Impartiality statement regarding citations. We, the editorial team, strongly encourage authors to cite the highest quality work believed to be most relevant to their article; we are impartial to the use of citations from Body Image versus other journals. We review and accept articles based on their scientific rationale, merits, design, analysis, and interpretation rather than the source of their citations.

Note regarding string citations. We encourage authors to avoid excessive string citations, whereby multiple citations support a single statement, finding, or proposition, when such citations would be superfluous. In many cases, one citation will suffice, and this citation should be the best supporting reference for that statement, finding, or proposition. All important previous work can be included, and if a cite is important, there often will be additional text that accompanies it. Please note that we are okay with the overall number of references.

Of note, the recommendation to avoid string citations does not apply to: Statements that include more than one finding. For example, "Over the past 10 years, researchers in a number of countries have begun to explore the relationship between positive body image and psychological well-being" needs multiple citations because authors are referring to researchers and countries (both plural. However, reference to all work that has explored this relationship is probably not needed.

As another example, "research shows that body dissatisfaction is correlated with disordered eating, anxiety, and depression" may include multiple citations, with different citations supporting different aspects of this statement. Systematic reviews and meta-analyses whereby the citations are linked to relevant themes/data that are included in the analysis.

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Coon, E., Berndt, M., Jan, A., Svyatsky, D., Atchley, A., Kikinzon, E., Harp, D., Manzini, G., Shelef, E., Lipnikov, K., Garimella, R., Xu, C., Moulton, D., Karra, S., Painter, S., Jafarov, E., & Molins, S. (2020, March 25). *Advanced Terrestrial Simulator (ATS) v0.88 (Version 0.88)*. Zenodo. https:// doi.org/10.5281/zenodo.3727209.

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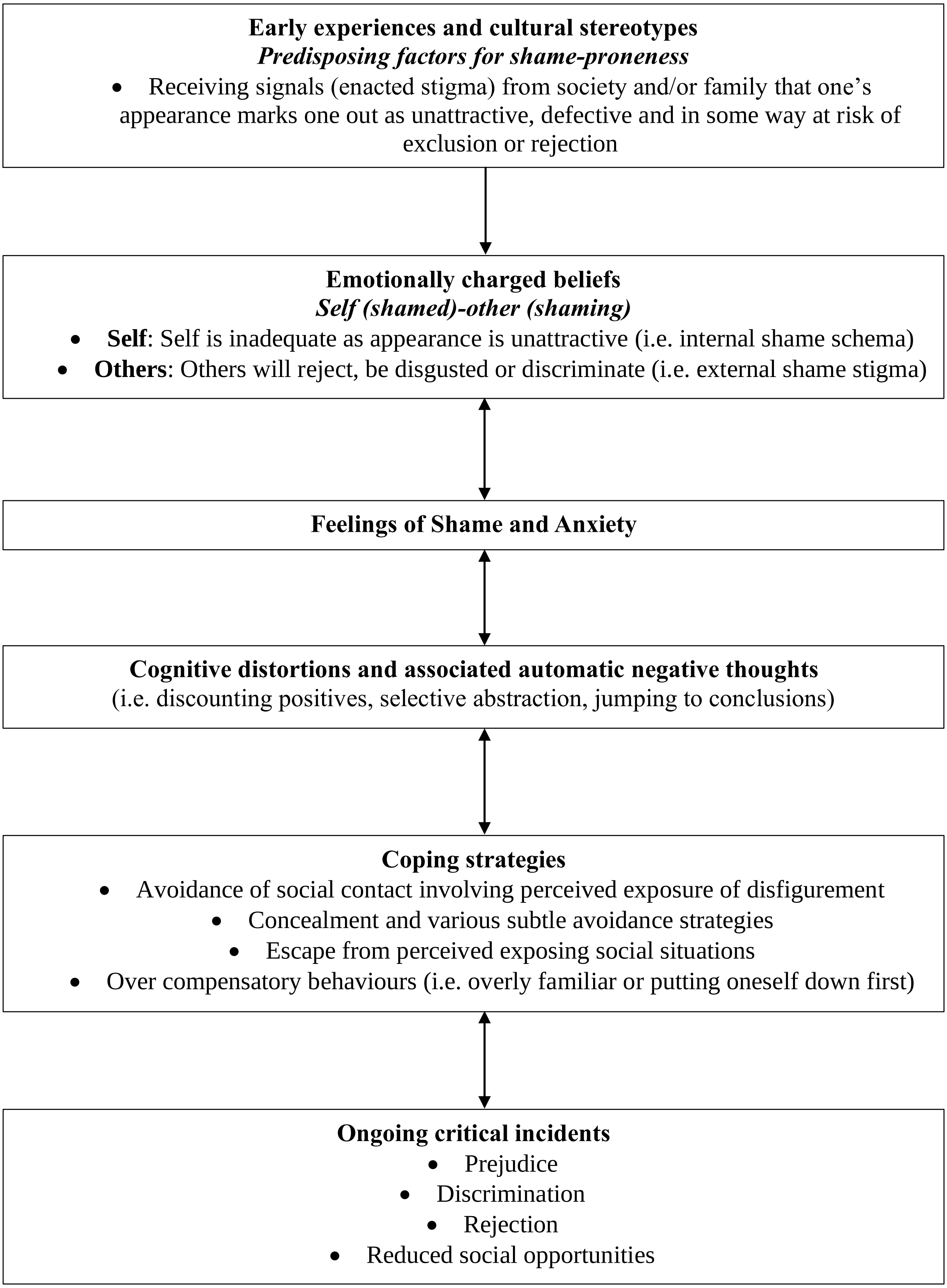
Appendix N

Model of Maintenance of Shame Proneness (Kent and Thompson, 2002)



Early experiences and cultural stereotypes

Predisposing factors for shame-proneness

- Receiving signals (enacted stigma) from society and/or family that one's appearance marks one out as unattractive, defective and in some way at risk of exclusion or rejection

Emotionally charged beliefs

Self (shamed)-other (shaming)

- **Self**: Self is inadequate as appearance is unattractive (i.e. internal shame schema)
- **Others**: Others will reject, be disgusted or discriminate (i.e. external shame stigma)

Feelings of Shame and Anxiety

Cognitive distortions and associated automatic negative thoughts

(i.e. discounting positives, selective abstraction, jumping to conclusions)

Coping strategies

- Avoidance of social contact involving perceived exposure of disfigurement
- Concealment and various subtle avoidance strategies
- Escape from perceived exposing social situations
- Over compensatory behaviours (i.e. overly familiar or putting oneself down first)

Ongoing critical incidents

- Prejudice
- Discrimination
- Rejection
- Reduced social opportunities

Appendix O

Central University Research Ethics Committee (CUREC) Approval Letter

MEDICAL SCIENCES INTERDIVISIONAL RESEARCH ETHICS COMMITTEE

Research Services, Boundary Brook House, Churchill Drive, Headington, Oxford, OX3 7GB
Tel: +44(0)1865 616575
ethics@medsci.ox.ac.uk

CONFIDENTIAL

Dr Ceara Moore & Miss Corah Lewis
Oxford Institute of Clinical Psychology Training & Research (OXICPTR)
Isis Education Centre
Warneford Hospital
Oxford

Dear Dr Moore and Corah,

Research Ethics Approval - CUREC 1 Ethics Approval Reference: R86293/RE001

Study title: Exploring the Impact of Perceived Social Support as a Moderator of the Relationship Between Public Stigma and Negative Outcomes in Facial Palsy

Short title: Stigma and Perceived Social Support in Facial Palsy

The above application has been considered on behalf of the Medical Sciences Interdivisional Research Ethics Committee (MS IDREC) in accordance with the University's procedures for ethical approval of all research involving human participants.

I am pleased to inform you that, on the basis of the information provided to the IDREC, the proposed research has been judged as meeting appropriate ethical standards, and approval has been granted for a period of **18 months**, commencing on **9th May 2023.**

Amendments

Should there be any subsequent changes to the study, you should submit details to the MS IDREC for consideration and approval. Details of changes must be listed on an amendment form.

Yours Sincerely

Mrs Leah Butts
Research Ethics Administrator

for

Dr Helen Barnby-Porritt Research Ethics Manager

Appendix P

Perceived Stigmatisation Questionnaire (Lawrence et al., 2006), reproduced with permission from first author

How do other people treat you?

During your normal day, you probably see and talk to many different people. We want to know how often people act in certain ways towards you.

For each question, rate how often people do certain things. Make your ratings about treated you **over the last year**.

Use the scale provided, *never* (1) to *always* (5). Circle your answers.

Never	Almost never	Sometimes	Often	Always
1	2	3	4	5

1. People are friendly with me.	1	2	3	4	5
2. People call me names.	1	2	3	4	5
3. People avoid looking at me.	1	2	3	4	5
4. People I don’t know act surprised or startled when they see me.	1	2	3	4	5
5. People are nice to me.	1	2	3	4	5
6. People don’t know what to say to me.	1	2	3	4	5
7. People I don’t know say “Hi” to me.	1	2	3	4	5
8. People laugh at me.	1	2	3	4	5
9. People are relaxed around me.	1	2	3	4	5
10. People feel sorry for me.	1	2	3	4	5
11. People pick on me.	1	2	3	4	5
12. People I don’t know smile at me in a friendly way.	1	2	3	4	5

13. People don’t know how to act around me.	1	2	3	4	5
14. People do “double takes” or turn around to look at me.	1	2	3	4	5
15. People are kind to me.	1	2	3	4	5
16. People bully me.	1	2	3	4	5
17. Strangers are polite to me.	1	2	3	4	5
18. People make fun of me.	1	2	3	4	5
19. People I don’t know stare at me.	1	2	3	4	5
20. People treat me with respect.	1	2	3	4	5
21. People seem embarrassed by my looks.	1	2	3	4	5

Appendix Q

Multidimensional Scale of Perceived Social Support (Zimet et al., 1998), reproduced with permission from first author

	Very Strongly Disagree	**Strongly Disagree**	**Mildly Disagree**	**Neutral**	**Mildly Agree**	**Strongly Agree**	**Very Strongly Agree**
1. There is a special person who is around when I am in need.							
2. There is a special person with whom I can share joys and sorrows.							
3. My family really tries to help me.							
4. I get the emotional help and support I need from my family.							
5. I have a special person who is a real source of comfort to me.							
6. My friends really try to help me.							

7. I can count on my friends when things go wrong.							
8. I can talk about my problems with my family.							
9. I have friends with whom I can share my joys and sorrows.							
10. There is a special person in my life who cares about my feelings.							
11. My family is willing to help me make decisions.							
12 I can talk about my problems with my friends.							

Appendix R

Pearson’s Correlations

	Age	Months with BP	Noticeability to you	Noticeability to others	Expected recovery	PSQ	MSPSS	PHQ	GAD	Combined distress score
Age	-	.241*	-	-	-	-	-.293**		-.205*	
Months with BP	.241*	-	-	-	-.430**	.224*	-	-	-	-
Noticeability to you	-	-	-	.722**	-	.254*	-	-	-	-
Noticeability to others	-	-	.722**	-	-	.334**	-	-	-	-
Expected recovery	-	-.430**	-	-	-	-	-	-	-	-
PSQ	-	.224*	.254*	.334**	-	-	-.346**	.340**	.285**	.331**
MSPSS	-.293**	-	-	-	-	-.346**	-	-.220*		-.205*
PHQ-8	-	-	-	-	-	.340**	-.220*	-	.780**	.941**
GAD-7	-.205*	-	-	-	-	.285**	-	.780**	-	.946**
Combined distress score	-	-	-	-	-	.331**	-.205	.941**	.946**	-

* Correlation is significant at the 0.05 level (2-tailed)

** Correlation is significant at the 0.01 level (2 tailed)