Exploring decision-making in paediatric surgery: A focus on the transition period and healthcare professionals' understanding of psychosocial and cultural factors in cleft orthognathic surgery.[1]

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Abstract

Objective: Previous research has explored the decision-making experiences of young people and their families in relation to orthognathic surgery (OS); however, less is known about how clinicians experience this process. Psychological, social, and cultural factors are highlighted when discussing young people's experiences of living with and seeking treatment for a cleft lip and/or palate (CL/P), however the literature suggests that clinicians often do not receive training in this. The thesis portfolio aims to identify barriers and facilitators to engaging in shared decision-making (SDM) in acute paediatric surgery, whilst exploring how clinicians understand and consider psychosocial and cultural factors, when supporting young people with CL/P in their decision-making for OS.

Design: A systematic review was conducted which explored the perceived barriers and facilitators to engaging in SDM in acute paediatric surgery. In supplement, a qualitative study was conducted with nine clinicians working in CL/P services which explored clinician awareness and consideration of psychosocial and cultural factors, and ethical dilemmas arising during SDM.

Results: The review revealed four key themes; understanding the patients' information needs, engaging in effective communication, promoting access to support and involving the patients support network. In focussing on OS, themes centred around the SDM environment, balancing patient autonomy and beneficence, and fostering opportunities for team liaison. Health inequalities were also highlighted, relating to socioeconomic status, financial and educational/employment considerations.

Conclusions: A complex interplay of factors were found to influence the decision-making process. Further research may wish to evaluate the effectiveness of SDM interventions, when supporting young people and their families. When focussing on OS, the gravity of making a decision is acknowledged whereby it is important to optimise the SDM environment and create opportunities for liaison. It is recommended that further research focus on exploring and addressing health inequalities.

Key words: Cleft, Surgery decision-making, Barriers, and facilitators.

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Acknowledgements

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Chapter One: An introduction to the thesis portfolio

A cleft lip and/or palate (CL/P) is a gap or split in the upper lip and/or palate, occurring in the first few weeks of craniofacial development in the womb. From the perinatal period onwards, patients follow a 20-year treatment pathway and can experience many challenges including difficulties with, feeding, hearing and speech. Surgeries to repair a CL/P typically occur during the first year of life, and many young people will require additional surgical procedures later in life (e.g. secondary speech surgery, alveolar bone graft, lip and/or palate repairs).

At the transition period (from childhood into adulthood, often around the ages of 16-18 years in cleft care), patients may elect for surgery to re-align the jaw, known as orthognathic surgery (OS). During this phase of treatment, patients are seen by a specialist multi-disciplinary team (MDT), consisting of Orthodontists, Surgeons, Speech and Language Therapists and Clinical Psychologists. Following surgery for OS, further surgery may also be elected for, including rhinoplasty and lip revision surgery.

The 20-year treatment pathway can be experienced as a difficult, and burdensome process for many young people, both in the sense of experiencing physical burdens related to pain, discomfort and having to make frequent trips to hospital, but also psychologically in the sense that many young people report experiencing accentuated feelings of being ‘different from others’ (Alansari et al., 2014). Alansari et al. (2014) in exploring how patients with CL/P experience the treatment pathway, illustrated through patient quotes how this process requires taxing perseverance which can be experienced as ‘fatiguing’ and ‘frustrating’. Patients also described how their interactions with clinicians could at times be solely focussed on the technical aspects of the treatment, without attention being paid to personal aspects of the patients lives. Furthermore, patients also reported that clinicians may address their parents, rather than them (young person), directly, which could result in the young person feeling that they lacked importance or control. Contrastingly, patients recounted occasions whereby clinicians did address them directly, which resulted in the patients feeling worthy and in control (Alansari et al., 2014).

These findings are also reported, when looking specifically at patients experiences of the transition period, whereby young people have reported feeling “ill-equipped” to make treatment decisions, experiencing clinicians to talk over them, or leave the young person out of discussions (Wogden et al., 2019).

NICE guidance (2016) outlines recommendations for transition planning, and for support before and after transition from child to adult services, advising that transition should consider the young persons capabilities, needs, and hopes for the future. It also suggests that clinicians consult with the young person about how and to what extent, they want their guardians to be involved in their transition to adult care. Part of this, may include supporting young people to develop their confidence in working with adult services by providing opportunities to raise any concerns and queries separately from their guardian (NICE, 2016). This may however, be dependent on the set up of each hospital/care provision and whether paediatric and adult services are separated in this way.

In assessing whether NICE guidance reflects young people’s experiences of transitioning from child to adult Cl/P services, the evidence base suggests that further consideration is needed regarding how these guidelines are implemented effectively, with recommendations being made around introducing transition workers, providing access to medical history documentation, increasing liaison between professionals and developing age appropriate resources to facilitate the transition process (Mcwilliams et al., 2023).

Health Inequalities and Cultural Beliefs in accessing Cleft Care

Health inequalities are defined by NHS England, as “unfair and avoidable differences in health across the population, and between different groups within society” (NHS England, 2022). Consequently, specific groups and communities, are more likely to experience barriers to accessing healthcare, resulting in adverse health outcomes. The reasons for this are complex, and include (but are not limited to), the availability of services within different areas, access to transport and childcare, language and literacy skills, receiving misinformation, experiencing stigma, and poor experiences of accessing care and services in the past.

With regards to cleft care, families of children and young people with CL/P who come from lower socio-economic groups, were found to have higher rates of failure to present to initial and follow-up clinical appointments (Smillie et al., 2014). Stock et al. (2016), in reporting on adult narratives of growing up with a Cl/P and factors influencing psychological adjustment, highlight socioeconomic status, culture and religion as background factors. For example, in discussing socioeconomic status and the impact this had on one patients’ experience of cleft care, a participant commented on how their childhood environment had an impact on their psychological adjustment to cleft. The financial implications associated with taking the bus back and forth to appointments was also highlighted, resulting in this patient missing appointments.

Delays in receiving treatment for CL/P repair are also reported to be greater for patients who identify as being from a marginalised background or for whom English is not their primary spoken language (Abbott et al., 2011; Zaluzec et al., 2019). Although there is little research describing how to address these disparities and systemic inequalities, the evidence base suggests that practises may be improved by increasing sharing of information amongst members of the cleft team and providers working in a more synchronised manner (Wagner et al., 2021).

Different cultural perspectives on the aetiology and management of clefts are also reported (Hasanuddin et al., 2023) with treatment perspectives encompassing herbal and animal remedies amongst South African communities to placing the child/young person in sand and exposing them to direct sunlight in some Indian communities. These practices stem from aetiological beliefs that a cleft results from infection, consuming “wrong foods,” medication or beliefs that the cleft is predestined by God, or resulted from supernatural/mystical forces (Hasanuddin et al., 2023). In some cases, families may opt for no intervention, as the cleft is viewed as being a gift from God or spiritual interventions and traditional medication may be sought, leading to adverse consequences. Awareness of these cross-cultural beliefs amongst clinicians, is therefore crucial in promoting effective collaboration and positive health outcomes.

The impact of experiencing a cleft, in respect to culture and religion, is also highlighted in a paper by Stock et al. (2016) in which a participant spoke of their experience of stigma and feeling less accepted in their community. The participant remarked on how being born with a CL/P impacted on them finding a marriage partner, mentioning that their facial scars meant that other families would not feel that they were a suitable partner for an arranged marriage, and that other factors, such as their education and family background, therefore became irrelevant (Stock et al., 2016). This further highlights the importance of clinicians understanding the social and cultural contexts and experiences of their clients.

Current understanding of psychosocial factors and motivations for surgery

The American Psychological Association (APA, 2018) defines psychosocial factors as "social, cultural, and environmental phenomena and influences that affect mental health and behaviour. These influences include social situations, relationships, and pressures, such as competition for education, health care, and other social resources; rapid technological change; work deadlines; and changes in social roles and status'". Psychosocial factors, experienced by clients with CL/P are reported widely across the literature with clinical guidelines stating that it is the responsibility of MDTs, to be sensitive to linguistic, psychosocial, economic, ethnic and physical factors that may affect dynamics between the team, patient and their family (The American Academy of Paediatric Dentistry, 2022).

In exploring psychological factors, research has found depression and anxiety to be reported around twice as often by clients affected by a CL/P, compared with controls, with psychological factors being associated with appearance related concerns and a desire to undergo further treatment (Ramstad et al., 1995).

Motivations for undergoing OS amongst patients, are reported to include improving facial and dental aesthetics and improving interpersonal relationships, psychological well-being, and self-esteem. In interviewing patients about their decision-making for elective surgery, improvement in self-perception was noted as the major goal, with physical and functional benefits also being described (Alansari et al., 2014). In contrast however, patients may hold the belief that undergoing OS will improve their romantic relationships or may lead to professional growth, overestimating the impact that surgery may have (Miguel et al., 2014). It is therefore of importance for MDT professionals to explore motivations and expectations around surgery, so that these are not misaligned (Miguel et al., 2014).

Although function and aesthetics have been noted as primary motivators for OS, research has challenged this, suggesting the reasons for seeking treatment are multifaceted, complex and may be subject to socio-cultural influences (Patcas et al., 2017). The authors highlight the need for clinicians to acknowledge and understand the socio-cultural context of their patients, recognising the influence that different geographic locations may have on motivations and expectations around surgery (Patcas et al., 2017).

Although it is recommended that MDTs should have an awareness of psychosocial and cultural factors, research has found that clinicians often do not receive formal training in this area. In particular, in a study conducted by Stiernman et al. (2019) exploring parental and clinicians views on psychosocial and education outcomes in patients with CL/P, no clinician reported having formal training on psychological factors. This sample included Surgeons, Orthodontists, Nurses and Speech and Language Therapists. Clinicians in the study, alternatively disclosed that their main source of information had been derived from seminars or conferences. It is therefore of importance to understand how MDT professionals working in cleft settings, identify and understand psychosocial and cultural factors and implement this knowledge when supporting young people, of which previous research has recommended conducting future research to explore clinician perspectives (Acum, 2018; Safarikova, 2021).

Ethical Dilemmas experienced during surgery decision-making

The American College of Surgeons (ACS) outlines six core ethical dilemmas that surgeons are presented with in their practice, with modern day practising surgeons identifying with the following four categories: ‘professional obligations,’ ‘competition of interests’, ‘truth telling' and 'end of life care'. In considering ethics applied to cleft care and surgery decision-making, the concepts of autonomy, beneficence, nonmaleficence and capacity have been debated (Strauss, 2002). Autonomy suggests that a patient is able to make decisions about their treatment, whereas in contrast, beneficence would imply that the professional provides what is felt to be in the patient's best interest. These principles therefore sit in contrast to one another, raising ethical considerations as to how clinicians navigate this dynamic and promote shared decision-making in a way that achieves favourable health outcomes.

An additional factor to consider, is the idea of competence and the age by which patients make autonomous decisions. Although the legal age a patient can consent to treatment is 16, children under the age of 16 are able to consent to their own treatment if they are believed to be Gillick Competent. In the case of craniofacial surgery, it may be considered "ethical" to empower a young person under the age of 15 to make decisions and contribute to discussions surrounding their care but may not be "ethical" to perform a procedure. This therefore poses considerations as to how these ethical dilemmas are navigated by clinicians working in cleft settings when considering the transition period of responsibility for the decision-making (from parent to young person) and OS in particular.

It is also of importance, to consider a patient's comprehension level, cultural context, educational background, and language skills when discussing consent, and is paramount that information is not only provided, but is understood (Kleinman, 1979). Particular attention should therefore be given to communication and any differences that exist between professionals, families, and patients they serve in ensuring informed consent.

Team working and communication

Health Education England (HEE), in producing the ‘Multidisciplinary Team Toolkit,’ define an MDT as “a team consisting of individuals drawn from different disciplines who come together to achieve a common goal” (HEE, 2021). The Social Care Institute for Excellence, further expand on this definition, outlining a set of aspirations for MDTs, regardless of the goal (SCIE, 2022). This includes, bringing together team members from diverse backgrounds, understanding each members roles and responsibilities in creating a shared identity, enabling better communication, productivity and trust within the team, and working in a holistic and personalised way in ensuring fewer errors are being made. The SCIE, also outline factors that may increase the effectiveness of MDTs, which include having a clear purpose, a leadership style that encourages contributions from different team members, having collaborative spaces and operating in a person-centred way.

One of the outcomes from the Clinical Standards Advisory Group report (Sandy et al., 1998), advised that having multidisciplinary teams delivering cleft care, would improve the outcomes for children born with CL/P. Within cleft MDTs, the importance of having clearly defined roles and responsibilities is also highlighted (Young, 1998; Sloper, 2004; Arskey et al., 2007; Choi and Pak, 2006).

Reassuringly, research exploring the perceptions of team members working in cleft services in the UK, has found clinicians to report positive perceptions of the way their teams work, with ‘team identity’ being the highest scoring area (Scott et al., 2015). This suggests that overall, individual team members have adopted a positive view of their role within the team. In contrast, the areas that received the lowest scores, concerned team foundations and leadership, which may warrant further exploration.

In reporting on the ‘ideal multidisciplinary cleft lip and palate care team,’ Frederick and colleagues (Frederick et al., 2022), reported that successful leaders led by consensus of their team members, and not in an authoritarian manner (Kummer, 2018). Additionally, the importance of working in a family-centred and collaborative way, in which input from each team member is gathered, is highlighted (Frederick et al., 2022).

Shared decision-making

Previous research has highlighted the role of shared decision-making (SDM) in relation to making decisions about elective surgery and has discussed models of decision-making implemented within healthcare settings (Acum; 2018). Citing a systematic review by Boss et al. (2016), Acum (2018), makes reference to SDM, and the impact of incorporating SDM practices on improving the quality of decisions made around elective surgery. Despite SDM being reported to have a positive impact on the quality of decisions being made, research has found that clinicians may underestimate how involved patients and their families wish to be in the decision-making process, which may hinder SDM. Acum (2018) also cites results from a thematic synthesis, which highlights reported barriers and facilitators to SDM, this including, acknowledging the expertise, values, and preferences of their patients (Barry et al., 2012) and creating awareness that patients can influence the decision-making process.

When specifically thinking about cleft settings, having insufficient understanding of facial difference and treatment and lack of involvement in decision-making relating to age and influence of ‘powerful’ stakeholders ‘taking charge’ were reported to be barriers to shared decision-making and in particular, autonomous decision-making (Bennett et al., 2019).

Facilitators to shared decision-making, reported in relation to cleft care, include collaborating with patients to find a mutually agreed upon treatment plan, and providing opportunities for patients to share their hopes and worries about treatment. It is reported that appropriate SDM, ensures the young person feels equipped to make decisions about their care and that they are knowledgeable about all the options available to them (Mcwilliams et al., 2023).

Park and Cho (2017), through the process of conducting a concept analysis, present a model for understanding paediatric shared decision-making, defining SDM as “the active participation of parents, children and health professionals in reaching a compromise via collaborative partnership with a common goal for their child’s health” (P482). The model is broken down into three components, ‘Antecedents’, ‘Attributes’ and ‘Consequences’, of which ‘making a compromise’, ‘collaborative partnership’, ‘the active participation of parents, children and HCPs’ [in SDM] and sharing a common goal for child health were identified as being attributes of shared decision-making in the paediatric field (P483). As a result, the researchers propose that this decreases decisional conflict, enables mutual empowerment, improves child health status, and has overall implications for the quality of paediatric health care. Making or ‘reaching a compromise’ is defined as reaching an ‘outcome via mutual agreement,’ of which the researchers describe the interaction between clinicians and patients as a ‘negotiation’ that should result in an agreement of joint decision being made. The thesis portfolio will discuss the findings, in relation to Park and Cho’s (2017) model of shared decision-making.

Chapter Two

Systematic Review

Title: what are the reported barriers and facilitators to shared decision-making related to surgery in paediatric settings? – A systematic review.

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Prepared in accordance with the requirements for submission to the Journal of Pediatric Health Care (see guidelines in Appendix 1)

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Abstract

Objective: To identify and report on the perceived barriers and facilitators to engaging in shared decision-making (SDM) for surgery within acute paediatric hospital settings, as reported by clinicians, children and young people and their families.

Design: A systematic literature search was conducted using three electronic databases, CINAHL, PsycINFO and SCOPUS. The search was conducted from January 2012 to April 2023, identifying studies that reported on barriers and or/facilitators to engaging in shared SDM in a paediatric surgical context.

Results: Seventeen papers were included in the final report, 12 of which included qualitative methodology or a component of. The remaining five papers included observational, mixed-methods, cohort and case study approaches with one scoping review being included. Four analytic themes and 11 subthemes were identified through the process of conducting a thematic synthesis. Analytic themes included understanding the patient's information needs, engaging in effective communication, promoting access to support, and understanding and involving the family's support network. Underpinning these themes, were sub-themes centring around physician language and communication, the provision of information, incorporating the values and beliefs of patients and families within SDM and considering the time and frequency of discussions, supporting the recommendations outlined in NICE guidelines (NICE, 2021).

Conclusions: The present review contributes to the literature highlighting barriers and facilitators to engaging in SDM and has implications for physician and patient interventions. It is recommended that future research evaluates the effectiveness of SDM interventions when focussed on paediatric surgical populations. Furthermore, it may also be beneficial, to explore how these skills and techniques, may need to be modified for paediatric settings.

Registration: CRD42023395334

Key words: Shared decision-making, paediatric, surgery, child and adolescent, barrier, facilitator, surgery decision-making.

Introduction

"Shared decision-making" (SDM) is defined by the National Institute for Health and Care Excellence (NICE) as "a collaborative process that involves a person and their healthcare professional working together to reach a joint decision about care" (P32), (NICE, 2021). NICE guidance suggests that this should be based on evidence, the individual's preferences, beliefs, and values and that the individual should understand the risks, benefits and possible consequences of different options that are presented to them. SDM is essential to delivering universal personalised care, of which the NHS Long Term Plan and NICE guidance both acknowledge how this involves considering the individual's strengths and needs and empowering patients to make decisions that are right for them. The importance of providing patients with the opportunity to choose to what degree they want to engage in decision-making, is also stressed (NHS England, 2019; NICE, 2021).

Although research has previously found clinicians to believe their patients prefer not to be involved in decision-making, or that they are not able to take an active role in this (Wogden et al., 2019; Hoffmann et al., 2014) a survey by the Care Quality Commission (CQC, 2020) and the results of a GP Patient Survey (2022) found that individuals want to be more involved in making decisions about their health and care; warranting further exploration in this area.

Much research has been conducted into the barriers and facilitators to engaging in SDM, with research focussing on the perspectives of clinicians, patients, and their families.

For the purposes of this systematic review, the term 'barrier' was defined as any factor that was perceived to obstruct successful implementation of SDM and the term 'facilitator' was understood as any factor that was perceived to help enable successful implementation of SDM.

Waddell et al. (2021) in reporting on the barriers and facilitators to implementing SDM from multiple perspectives, discussed system level factors such as having a lack of guidance or training around SDM in addition to clinician-related factors impacting on SDM such as holding beliefs that the patient or colleagues would not want to engage in SDM, lack of training and assuming the patient understands the information given. Patient related factors included not being provided with adequate information to be able to make a decision, provider biased information, limited understanding of risk or knowledge about their condition and options, perceived ‘unacceptability’ of asking clinician questions and believing that the clinicians role is to make the decision (Waddell et al., 2021). In examining facilitators to engaging in SDM, these were described as including the patient in SDM as soon as possible, involving family, having a trusted relationship with the clinician, including SDM in medical students training and including SDM within professional role descriptions for clinicians.

In looking at paediatric settings in particular, which for the purpose of this review, are defined as settings whereby clinicians specialise in the medical or surgical care of children and young people up until the age of 18, similar barriers and facilitators have been reported to those identified in the adult literature. These include, receiving poor quality information about the condition and/or treatment options that were not appropriately tailored to the child and family’s health literacy needs. Trust and respect between the clinician and family was also reported to be a key facilitator to engaging in SDM (Boland et al., 2019).

The legal age of consent to treatment, is 16, however a young person under this age may be considered able to make decisions about their care, if they are perceived to be 'Gillick Competent'. Effective shared decision-making, should therefore aim to empower young people, especially when working with young people who are expected to transition to adult services (NICE, 2016). Particular attention should also be drawn to the values and belief systems of families, and the social and cultural contexts of which they derive.

The present review

The current review aims to identify and report on the perceived barriers and facilitators to engaging in SDM, with a particular focus on SDM applied to surgery decision-making in paediatric settings. In synthesising the literature, the current review aims to answer the following question; 'what are the perceived facilitators to engaging in shared decision-making when making decisions about surgery within paediatric settings?'.

Definitions of terms

In making reference to SDM throughout this review, the NHS England definition of shared decision-making was used. NHS England define shared decision-making as follows; "a collaborative process through which a clinician supports a patient to reach a decision about their treatment. The conversation brings together the clinician's expertise, such as treatment options, evidence, risks, and benefits, what the patient knows best: their preferences, personal circumstances, goals, values and beliefs" (NHS England, 2019).

Methods

Design

A systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. A protocol outlining inclusion and exclusion criteria and process for carrying out the systematic review was developed and registered on the international prospective register of systematic reviews (PROSPERO; 2023 CRD42023395334).

Information sources and search strategy

Included papers were identified through searches carried out on the following databases: PsycINFO, SCOPUS and CINAHL. Guidance was sought from an experienced university librarian in deciding upon the search strategy, in particular, with identifying search terms and time period for the search. Search terms focussed on the setting where SDM was carried out, the population being studied and using similar terms to encompass ‘barriers’ and ‘facilitators’. The final search terms and inclusion and exclusion criteria are presented in Tables 1 and 2. A previous systematic review explored the literature surrounding patient-reported barriers and facilitators to shared decision-making in healthcare up to 2012 (Joseph-Williams et al., 2014); therefore a time period of January 2012 to April 2023 was chosen to ensure the literature was contemporary.

In the UK, a child or young person may remain under the care of their Paediatrician, up until their 18th birthday (NHS England, 2017) although often young people may transfer from age 16 depending on the service and health condition guidelines. The age of consent to treatment is 16, however if a young person is deemed to lack capacity, a parent or guardian may be required to make a decision in their best interest. Encompassing the literature up to the age of 18, may therefore provide insight into any differences reported with respect to perceived barriers and facilitators to SDM, occurring between the ages of 16 and 18, that otherwise may have been excluded. For this reason, the current review includes papers that discuss SDM in relation to children and young people aged 18 years or below.

Table 1

Inclusion and Exclusion Criteria for systematic review

Inclusion Criteria	Exclusion Criteria
Written in English	Not written in English
Dated between January 2012 and April 2023	Dated before 2012 or after April 2023
Makes reference to factors that obstruct or enable successful implementation of shared decision-making (Barriers and/or Facilitators)	Population aged 19+ (Adult) or where the paper discusses both adult and child populations within the same paper.
Makes reference to shared decision-making in the context of surgery	Barriers and/or facilitators discussed but not in the context of surgery decision-making
Shared decision-making discussed in the context of paediatric setting (aged 18 years of below).	Paper reviews a decision-making tool or decision aid/communication aid and does not discuss barriers and or facilitators to shared decision-making.
	Where barriers and or/facilitators to engaging/implementing shared decision-

	making are discussed in the context of treatment planning only and not surgery.
	Paper outlines a protocol or is an evaluation of guidance/policy.
	Focusses on the perspective of one group in isolation, not shared decision-making (e.g. discussing parent reported factors, but not shared decision-making that involves another party).

Table 2

Search Terms for literature review

Concept 1	**Concept 2**	**Concept 3**	**Concept 4**
Decision Mak*	**Child OR Adolescen***	**Surg***	**Hospital**
OR Patient Participation	OR Teen*	OR Operation	OR Acute Setting
OR Patient Involvement	OR Young Person*		OR Hospital Setting
OR Informed Decision	OR Juvenile		OR Healthcare
OR Informed Choice	OR Paediatric		OR Healthcare Facility
OR Choice Behaviour	OR Pediatric		OR Inpatient

OR Decision Support	OR Youth		OR Inpatient Setting
			OR Clinic
			OR Ward

Search results and study Selection

3367 articles were identified through the search strategy of which 3045 articles were screened by their title and abstract. In the initial phase, titles and abstracts were screened by two reviewers (HC and SC) against the pre-defined inclusion and exclusion criteria (Table 2), prior to carrying out full text searches. 2993 articles were removed in the initial phase, as they did not meet the inclusion criteria, or were identified as being additional duplicates. The second reviewer (SC) screened 7% of titles and abstracts during the initial phase in which screening was carried out using 'Rayaan', a systematic review tool. Rayaan allowed reviewers to be blinded to each other's screening decisions of which any discrepancies were later discussed against the eligibility criteria in reaching a consensus. Following this, full-text articles were screened by HC for eligibility and inclusion in the final report, 20% of which were also screened by a secondary reviewer (RU). Any disagreements were resolved in which 17 papers were included in the final report. Figure 1 provides a breakdown of the literature search, illustrated by a PRISMA flow diagram, (see Figure 1).

Data extraction and analysis

The following information was extracted from included full-text papers:

- Author, year paper was published and country where research was based.
- Title of paper
- Age of child/population being studied.
- Type of surgical procedure or condition being studied.
- Participants/stakeholders who reported the barriers and/or facilitators to engaging in shared decision-making.
- What barriers and/or facilitators were being reported.

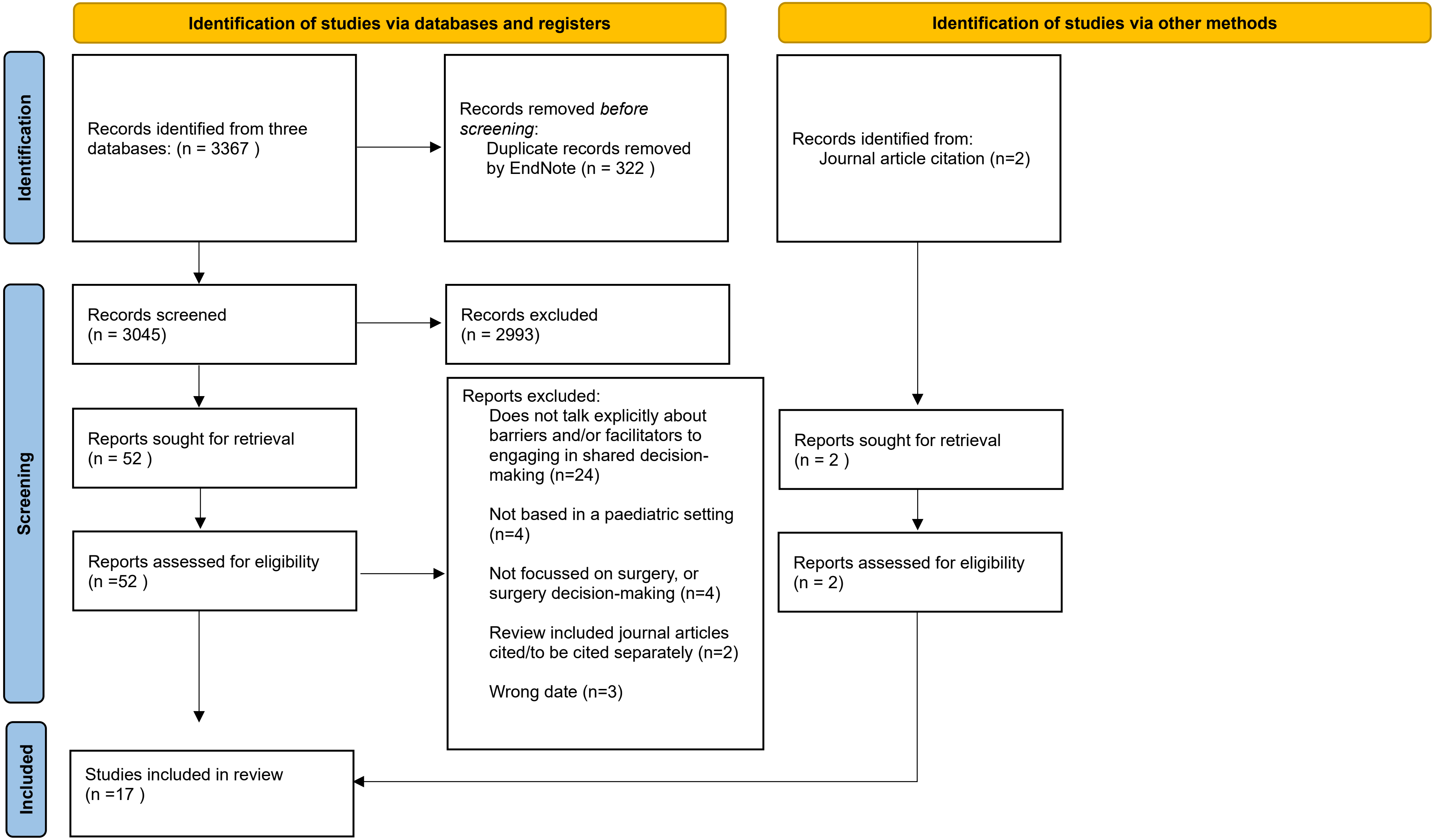

Figure 1: *PRISMA Flow Diagram displaying the screening and study selection process*

Results

Identified studies and characteristics

Of the 17 papers that were included in the final analyses and report, 12 of these papers had a purely qualitative methodology or component. Of the remaining five papers, one paper had an observational methodology, one paper was quantitative, one paper discussed case studies, one paper was a scoping review and another was a mixed methods study including both qualitative and quantitative components. Across papers, research was largely conducted in the USA (n=8) or the UK (n=7). Research was also conducted in Canada (n=1) and Sweden (n=1). Papers encompassed the perspectives of families, clinicians, and the young person themselves, with the age of the paediatric population being reported on, ranging from 1 day old to 18 years of age. The papers included clinicians and families, associated with various surgical specialities, including epilepsy surgery, neurosurgery, plastic surgery, orthopaedic surgery, cardiac surgery, and general surgery. Conditions being reported on, included tonsillectomy, fracture, shunt malfunction, hernia repair, transplant, dorsal rhizotomy amongst other conditions. (See Table 3 for further information).

Table 3

Study characteristics of included studies

Author, Year and Country	Title	Age of child/Population	Type of surgical procedure and/or condition	Participants reporting barriers and/or facilitators	Barriers and/or facilitators to SDM reported
Carlisle et al. (2023) USA	A valued voice: A qualitative analysis of parental decision-making preferences in emergent paediatric surgery	1 day old (Min) to 13 years old (Max)	Surgery for cancer, an emergent operation while in the neonatal intensive care unit (NICU) or extracorporeal membrane oxygenation (ECMO). **Conditions** 1. Wilms Tumour 2. Neuroblastoma 3. Persistent Pulmonary Hypertension 4. Congenital Diaphragmatic hernia 5. Sepsis 6. Bowel Perforation 7. Necrotizing Enterocolitis 8. Persistent Pulmonary Hypertension 9. Gastric Perforation	Parents	**Facilitators** 1. Surgeons providing recommendations – feeling more confident and involved in caring for their children. 2. Providing concrete scaffolding to guide the further pursuit of knowledge. 3. Providing information and statistics or descriptions of the surgical procedure 4. Slow conversational pace with significant repetition. 5. Opportunity to ask questions about their child's care. 6. Explaining the options 7. Viewing written and graphical information (drawings, brochures, figures) 8. Surgeons guiding online searches by providing reliable websites or preferred social media groups to parents. 9. Increased opportunity for virtual communication adjuncts (e.g. virtual presence during surgical rounds). 10. Providing parents with opportunities to advocate for their child as well as for families with similar surgical problems. **Barriers** 1. Repeated questions from multiple teams 2. Use of acronyms

					3. Poor surgeon demeanour
Atsaidis et al. (2022) Canada	Understanding the effectiveness of consent processes and conversations in pediatric surgery: A systematic-scoping review	<18 years (Paediatric)	1. Plastic surgery 2. ENT 3. Multi-Speciality 4. General Paediatric Surgery 5. Cardiac Surgery 6. Pediatric Urology 7. Ophthalmology 8. Orthopaedic Surgery 9. Neurosurgery	1. Patients and families 2. Surgeons and surgical trainees 3. Other clinicians (non-surgical physician or nurses) 4. Hospital administrators or professional policy makers.	**Facilitators** 1. Use of multimedia/visual tools (images, videos, presentations). 2. Providing written information 3. Repeat meetings and discussions with surgical team. 4. Individualising communication to specific patient/family. 5. Physician empathy 6. Trust in physician 7. Adequate time and opportunity for questions 8. Providing structure 9. Gauging parental comprehension 10. Considering moral values and beliefs of patients 11. Providing realistic goals 12. Disclosing the surgeon's level of expertise **Barriers** 1. Power imbalance – patients feeling intimidated or less knowledgeable. 2. Being provided with too much, or too little information. 3. Presence of added stress 4. Parental preoccupation with having a child in the room. 5. Limited language/comprehension skills.

Boss et al. (2017) USA	Parent experience of care and decision-making for children who snore.	Ages 2-17	Sleep-disordered breathing/Adenotonsillectomy	Parents	**Facilitators** 1. Responsiveness of clinicians (answered questions, remained supportive, available). 2. Physician disclosure of medical evidence or information (verbal and written form) – percentages. 3. Trusting relationship with paediatric clinician. 4. Confidence portrayed by surgeon. 5. Personal characteristics (Genuine, understanding, not rushed, welcoming). 6. Relationship with Paediatrician who made the referral and inclusion of Paediatrician in the consultation and treatment process. **Barriers** 1. Lack of information 2. Personal characteristics (arrogance, judgemental)
Samanta et al. (2022) USA	Physicians' Perspectives on Presurgical Discussion and Shared Decision-Making in Pediatric Epilepsy Surgery	Paediatric Epilepsy Centre Unspecified	Paediatric Epilepsy Surgery	Clinicians.	**Facilitators** 1. Going through the decision-making process again with families until a decision has been made, revisiting discussions. 2. Providing written information, handouts, charts, and digital tools (apps, websites, webinars, podcasts). 3. Patient testimonial videos suggested. 4. Surgeons discussing surgery statistics with family. 5. Suggestion of further physician specific education and training in effective communication, SDM and care planning.

					6. Use of social media posts to provide support, information and resources for potential patients and families. **Barriers** 1. Providing too much information – overwhelm 2. Further training required around communicating and tailoring evidence to patients and families. 3. Discussing statistics too early on.
Lecouturier et al. (2015) UK	Treating childhood intermittent distance exotropia: a qualitative study of decision-making	Children up to the age of 12 years.	Intermittent distance exotropia (X(T))	Clinicians Parents	**Facilitators** 1. Providing written information (clear and reliable information). 2. Having the opportunity hear from parents whose child had undergone the treatment. 3. Exploring values and preferences of families. 4. Including current data on success rates – discussing this with parents.
Lerret et al. (2016) USA	Parents' perspectives on Shared Decision-making for Children with Solid Organ Transplants	3 weeks to 17.5 years.	Heart, kidney, liver, lung, or multivisceral transplant.	Parents	**Facilitators** 1. Provider attributes (Knowledgeable, approachable, transparent, accessible, dependable, supportive) 2. Overtly discussing SDM. 3. Humility from clinicians – naming when they do not have 'all the answers' 4. Delivery of information – professional, considerate and in complete manner. 5. Bringing lists of questions or concerns to meetings. 6. Parents seeking information beforehand (Reading) 7. Willingness to talk with families until plan understood.

					Barriers 1. Conflicting information from what parents read or were told compared with information from the healthcare team. 2. Being made to feel inadequate, unintelligent or feeling judged by clinicians when asking questions.
Links et al. (2020) USA	Parental role in decision-making for pediatric surgery: Perceptions of involvement in consultations for tonsillectomy.	Aged 2-17	Tonsillectomy	Parents Healthcare professionals	**Facilitators** 1. Heightened information sharing/Information transparency. 2. Using clear, layman language 3. Tailoring decision-making roles to patients needs and fostering trust. 4. ‘Paternalistic communication style’ depending on needs. **Barriers** 1. Lower quality information 2. Clinician use of jargon 3. Parents desiring SDM but have limited involvement. Clinicians underestimating parents’ preferences to share decisions.
Papiez et al. (2021) UK	A qualitative study of parents’ and their child’s experience of a medical epicondyle fracture.	Age 7-14	Medial Epicondyle Fracture	Parents Children/ Young Person	**Facilitators** 1. Developing trusting relationships with surgeons 2. Preferring surgeons to make decisions. 3. Provision of consistent information 4. Family-centred approach that enabled parents and young people to develop the confidence to make a decision within a supportive environment.

					5. Answering questions 6. Openness to repeated visits. 7. Reassurance 8. Feelings of personally being cared for and focussing on child. 9. Surgeons appreciating and contextualising the importance of pre-existing experience and beliefs
Leu et al. (2021) USA	Assessment of Parental Choice Predisposition for Tonsillectomy in Children.	Aged 2 to 17	Tonsillectomy	Parents Children/Young Person Clinicians	**Facilitators** 1. Clinicians having an awareness of parent predisposition in effectively tailoring discussions. 2. Clinicians initiating discussions about treatment preferences and aligning with families' values. 3. Focusing discussions on treatment benefits to a child's particular clinical situation as well as the parents' unique concerns (individualising discussions) 4. Weight of the primary care clinician relationship and influence on decision-making. (e.g. Pediatriciians) **Barriers** 5. Parents who had a predisposition to choose tonsillectomy were less likely to engage in decision-making process, less likely to ask questions. 6. Perception of adequate knowledge amongst parents may hinder honest discussions between clinicians and parents and may prevent parents considering alternative treatment options.
Smith et al. (2013b)	Parents' experiences of living with a child with hydrocephalus: a cross	Ages 2-13	Shunt malfunction	Parents	**Facilitators** 1. Information delivered clearly and in a way that demonstrates empathy.

UK	sectional interview-based study.				2. Planned educational events, where there were opportunities to meet with other parents. **Barriers** 1. Parents perceiving clinicians as unwilling to engage in in-depth discussions. About their child's needs. 2. Restricting the information shared by parents. 3. Uncertainty around how to engage with clinicians and contribute to care decisions. 4. Information 'overwhelming' with overuse of complex medical terminology. 5. Emotions experienced on first learning about diagnosis can make it difficult for parents to comprehend information about their child's condition.
Smith et al. (2013c) UK	Are parents and professionals making shared decisions about a child's care on presentation of a suspected shunt malfunction: a mixed method study?'	Ages 1-15	Shunt malfunction	Parents Healthcare Professionals	**Facilitators** 1. Eliciting and valuing parental concerns 2. Recognising parental knowledge 3. Establishing rapport and continuity with professionals 4. Building effective and lasting relationships with parents 5. Listening – listening to patient story 6. Information sharing 7. Valuing parent experiences **Barriers** 1. Time constraints 2. Workload pressures 3. Lack of privacy when interacting with parents 4. Not feeling listened to 5. Being excluded when professionals grouped together (e.g. during wards rounds/care planning)

Waite et al. (2023) UK	'Wanting no regrets': Parental decision-making around selective dorsal rhizotomy.	Ages 4-11	Selective dorsal rhizotomy	Parents	**Facilitators** 1. Long-standing and trusting relationships with local clinicians. 2. Opinions, experience, and knowledge of the paediatrician 3. Experience-based knowledge of other parents: Information/outcomes/experiences from other parents who had been through SDR. 4. Suggestion of accessing psychosocial support to help make a decision away from medical clinicians. 5. Support from wider family. 6. Suggestion of bridging the gap between parents and health professionals on social media platforms. 7. Clinicians supporting information seeking 'and decision-making with respect and mutuality. **Barriers**. 1. Gaps in parental knowledge due to absence of long-standing and trusting relationship with local clinicians and short staffing. 2. Feeling intimidated/constrained/clinicians displayed apathy to engage in open dialogue. 3. Incongruency between own research and the narrative provided by clinicians. 4. Inconsistencies in online sources (e.g. unbalanced information).
Timmermans et al. (2018)	Does patient-centred care change genital surgery decisions? The strategic use of clinical	0 to 5.5 years	Genetic Surgery/Disorders of Sex	Clinicians caregivers	**Facilitators** 1. Clinicians encouraging parents to consult additional materials from the patient advocacy community.

USA	uncertainty in disorders of sex development clinics.		Development.		2. Clinicians making other families available for consultation. 3. Clinicians providing parents with information about support groups and outside information. 4. Clinicians actively soliciting questions or concerns from parents. 5. Clinicians emphasising that there is ‘no urgency’ and allowing time to make decisions. 6. Addressing anxieties from parents
Heath et al. (2016) UK	Putting children forward for epilepsy surgery: A qualitative study of UK parents’ and health professionals’ decision-making experiences.	2.5 to 18 years	Epilepsy Surgery	Parents Clinicians.	**Facilitators** 1. Information provided in lay language. 2. Adequate information about surgery presented in a variety of formats (Information packs, booklets, photographs, videos, email address for asking questions to inform FAQ). 3. Providing ‘frequently asked questions’ information. 4. Parents increasing own knowledge through seeking out additional information (online information and videos) – empowering. 5. Discussing treatment options with family members (partners, siblings, grandparents, child). 6. Early and softer introduction of surgery as possible treatment option. 7. Peer support – accessing the experience and expertise of other parents who had followed a similar treatment pathway. 8. Involving parents in decision-making from the out-set. 9. Formalizing families as part of the child's MDT by systematically incorporating patient/family perspectives within case presentations in MDT meetings.

					10. Eliciting and incorporating opinions and assessments from multiple disciplines. Working cohesively as a team towards a shared goal. **Barriers** 1. Inadequate information received from professionals. 2. Limited parental involvement in initial discussions regarding surgery candidacy 3. Not including patient-related information in MDT discussions. 4. Patient views not being sought until after the team have discussed treatment options.
Sjoberg et al. (2015) Sweden	The perspective of children on factors influencing their participation in perioperative care.	8 to 11 years	Tonsillectomy Adenoidectomy Teeth extraction Tenotomy Eye, Bulbus Oculi Outer ear plastic Achilles tendon extension	Children	**Facilitators** 1. Preparatory information from clinicians and family members (parents searching the internet for information and videos). 2. Being listened to/mutual interaction. Clinicians asking children how they are perceiving the situation and if they have questions. 3. Children being able to decide on 'small matters' e.g. holding breathing mask, starting anaesthesia, food choices after surgery. 4. Interpersonal qualities of clinicians (kind, available, reassuring). 5. Meeting clinicians who would be involved in the surgery. **Barriers** 1. Wanting more precise/more detailed information about the procedure.

					2. Preparatory information not being consistent with child's experiences/insufficient information.
Almoajil et al. (2023) UK	Exploring the factors that influence stakeholders' expectations and subsequent perception of lower limb orthopaedic surgical outcomes for ambulant children with cerebral palsy – a qualitative study.	8 to 18 years	Lower limb orthopaedic surgery/cerebral palsy	Parents, Clinicians Children/young person	**Facilitators** 1. Providing package of information, based on the individual child and family's needs. 2. Showing videos of post-operative changes (presenting best and worst-case scenarios). 3. Allowing sufficient time for decision-making and questions. 4. Shared goal setting – learning about goals from the child's perspective. Including care-givers goals within this. 5. Meeting other families as a motivator for children towards surgery and managing expectations. 6. Family encouragement, patients' self-determination, and previous experiences. **Barriers** 1. Not receiving much information 2. Lack of time and resources 3. Lack of continuous health services 4. Previous experiences and pre-operative anxiety. 5. Uncertainty about the unknown (children).
Loeff & Shakhsheer. (2021) USA	The ethics of informed consent and shared decision-making in pediatric surgery.	Unspecified, Paediatric.	Paediatric Surgery (case studies centring on the below) Neuroblastoma Gastroschisis	Authors reporting on physician and parent experiences.	**Facilitators** 1. Surgeon having a good understanding of the patients social and cultural background, aligning with the patients' values, beliefs, and expectations. 2. Unhurried, honest discussions. 3. Surgeons' skill in navigating dilemmas or conflicts in therapeutic choices and decisions. 4. Allowing time/space by modifying delivery of information.

5. Quantifying risks numerically and percentage reporting.
6. Providing supplementary written materials/simplifying information.

Barriers

7. Surgeons’ personal biases
8. Lack of consensus between the parents or between parents/caregiver and surgeon.
9. Emotional and psychological state of participants.
10. Language barriers

Critical appraisal

17 papers were appraised by the primary author (HC) in the first instance. Five papers were secondary appraised by the primary supervisor (KM) in order to reduce risk of bias. Findings were discussed between researchers and any disagreements resolved. Papers were appraised by six different quality appraisal tools, owing to differences in the methodology and design. The author consulted with an experienced librarian during the appraisal process, in identifying and assessing the suitability of different appraisal tools.

Qualitative studies were appraised using the Critical Appraisal Skills Programme (CASP) Checklist for Qualitative Studies (CASP, 2018) and quantitative research appraised using the CASP Checklist for Cohort Studies (CASP, 2018). The JBI Critical Appraisal Checklist for Systematic Reviews and Research Syntheses (JBI., 2020) was used in appraising scoping reviews, with papers reporting on case studies being appraised using the JBI Critical Appraisal Checklist for Case Reports (JBI., 2020). Observational studies were appraised using the National Heart, Lung, and Blood Institute (NHLBI) Quality appraisal tool for Observational and Cross-Sectional Studies (NHLBI, 2013) and mixed methods studies appraised using the Mixed Methods Appraisal Tool (MMAT) devised by Hong et al., 2018). A copy of the screening tools can be found in Appendices 3 to 8.

In assessing the quality of studies using CASP, computing an overall score is not advised (CASP, 2018; Noyes, 2018). This is also advised against when using the Mixed Methods Appraisal Tool (Hong et al., 2018). Appendix 2 therefore provides a table showing what criteria were met, in providing a discussion around the quality of papers using CASP and the MMAT. Half of the papers appraised using the CASP tool for qualitative studies met

90% of the CASP appraisal criteria. One paper met all criteria, and the remaining three papers met between 60 to 80% of the appraisal criteria (see Appendix 2).

In assessing the quality of papers using the JBI checklists, research has computed the percentage of criteria met in assessing quality (Valesan et al., 2021). Studies using the JBI tool were therefore assigned a quality rating of high risk of bias (<49% criteria met), medium risk of bias (50-69% criteria met), low risk of bias (70%+ criteria met) and the ratings for study's assessed using JBI are provided in Appendix 2. In using the NHLBI checklist, a rating system of 'good,' 'fair', or 'poor' is advised, which is based on a critical appraisal of study characteristics (e.g. dropout rate, allocation bias, variance in baseline characteristics). An overview of criteria met for studies using NHBLI are provided in Appendix 2.

Limitations resulting from Critical Appraisal

The main limitations noted across papers, concerned researchers critically examining their own role, potential bias, and influence during the research process. Regarding the appraisal of observational papers, it was noted that less than 50% of eligible participants, took part in the research with power, sample size and time periods not being stated (Leu et al., 2021), this paper was therefore given a rating of 'Poor'. In assessing the quality of mixed methods approaches (Smith et al., 2013c) limitations were noted with respect to the integration of qualitative and quantitative methods and illustrating the value of conducting a mixed methods study in the interpretation of the findings. Furthermore, although papers mentioned receiving approval from an ethics review board and REC, warranting a 'yes' in appraising the discussion of ethics, the discussion around ethical considerations generally did not expand this. An overview of the breakdown of criteria, can be found in Appendix 2.

Thematic Synthesis

A thematic synthesis was undertaken by the researcher, reporting on the barriers and facilitators to engaging in shared decision-making. This process followed the steps outlined by Thomas & Harden (2008). This methodology was chosen, as it allowed for the analysis of qualitative studies and identification of themes appropriate to the systematic review.

Data was firstly coded, capturing information relating to individual experiences and perceptions around the reported barriers and facilitators to engaging in shared decision-making. Secondly, similarities between codes were identified and codes grouped into 'descriptive themes'. This allowed for patterns to be identified across study data. The third stage involved synthesising findings across studies through the development of 'analytic themes', allowing for the interpretation of meaning applied to the research question. Four analytic themes and 11 sub-themes were identified through the thematic synthesis (See Figure 2).

Figure 2: *Diagrammatic representation of descriptive and analytic themes, depicting the perceived barriers and facilitators to engaging in SDM within Paediatric Surgical Settings.*

1. Patient Information Needs

1.1 Provision of information and Information seeking. 15 out of the 17 articles included in the review, made reference to the provision of information as either a barrier, or facilitator to engaging in SDM. Tailoring information to the child and family's needs, providing information in a range of formats and multimedia; and information being clear, reliable and consistent were all reported to facilitate SDM (Heath et al., 2016; Lecouturier et al., 2015; Papiez et al., 2021). Conversely, providing too much information that was felt to be overwhelming, not receiving enough information, or receiving information that is felt to be inadequate, not precise or detailed and misaligning with children's expectations, were experienced as barriers to engaging in SDM (Samanta et al., 2022; Atsaidis et al., 2022; Sjoberg et al., 2015; Heath at al., 2016).

Parents were also found to seek out additional information online in their attempt to increase their own knowledge, which was reported to be empowering (Heath et al., 2016). Furthermore, it was mentioned that surgeons may have a role in guiding online searches by providing reliable website and preferred social media groups (Carlisle et al., 2022). This may also reduce access to online information and sources that are considered by some parents to be unreliable and unbalanced (Timmermans et al., 2018). Waite et al., (2022), illustrated this as follows, presenting the following quote from a parent, "Facebook it is very opinionated and not necessarily right, so much misinformation on there". (p386). The role of clinicians in guiding online searches, was also put forward by Loeff and Shakhsheer (2021) as follows, "In some ways, the proactive patient can become more empowered with basic knowledge to engage in an informed consent dialogue, however, the surgeon may have to interpret data and explain misconceptions picked up by the patient" (p2).

1.2 Evidence base: providing statistics and percentages. As reported across papers, facilitators to SDM included surgeons discussing population-based surgery statistics with families and providing outcome data (Lecouturier et al., 2015; Samanta et al., 2022). It was further noted that quantifying risks numerically and percentage reporting, was a benefit within SDM discussions (Loeff & Shakhsheer (2021), "Percentage reporting may help a patient understand how often a complication happens in general and how likely it is to happen to their child." (p2). In contrast, discussing surgery statistics too early in the SDM discussion, however, was reported to be a barrier to engaging in SDM (Samanta et al., 2022).

1.3. Time and/or frequency of discussions and consultation. Being provided with adequate time to validate and address parental concerns, revisiting discussions, and openness to repeat visits in order to consider decision-making and answer questions, were acknowledged as enhancing the shared decision-making process (Samanta et al., 2022; Timmermans et al., 2018, Papiez et al., 2021). In contrast, time pressures/constraints and lack of resources were reported to hinder SDM (Almoajil et al., (2022) Smith et al., 2013c).

1.4. Surgeons providing recommendations. For some parents and families, it felt important that surgeons provided structure and recommendations within SDM discussions (Atsaidis et al., 2022; Carlisle et al., 2022). This was illustrated by Carlisle et al., (2022) through the following parent quotes, "ultimately the surgeon needs to share what they're recommending and how they want to proceed' (p534). Another parent explained that surgeons' recommendations "kind of gave me something to look up online to try to figure out [things]" (p534). Despite surgeon recommendations being perceived as helpful, in one case, Carlisle et al., (2022) reported that the recommendation was found to be unhelpful, due to the blunt manner in which this was delivered, suggesting barriers and facilitators to engaging in SDM, centred around communication and interpersonal characteristics.

2. Networks and Support Systems

2.1. Patient values, beliefs, and Cultural background. A key feature of SDM, involves considering patient preferences and values. Similarly, the importance of understanding the patient's social or cultural background has also been acknowledged as a facilitator to engaging in SDM, in the sense that the surgeon will be more prepared to recommend surgical treatments that align with the patient's values, beliefs and expectations (Loeff & Shakhsheer., 2021). Lecouturier et al., (2015), in discussing the importance of incorporating patient preferences and values into treatment decisions, present the following passage: "In some families there had been conflict between parents on the decision around surgical intervention and clinicians could have a role to play; exploring the understanding, values and preferences of both parents may have helped to facilitate decision-making and alleviate concerns.". (p9). This further emphasises the role clinicians have, in navigating SDM discussions, paying particular attention to patient preference, values and cultural background.

2.2 Relationships with/and involvement with other professionals. Families reported that the relationship with, and opinion, experience, and knowledge of their child's paediatrician, impacted on SDM around surgery, reporting the involvement of the paediatrician in consultations and treatment process to be a facilitator to SDM (Boss et al., 2017; Waite et al., 2022). Children who were interviewed about their experiences have also reported that it is helpful to meet the clinicians that would be directly involved in their surgery beforehand, when making decisions in the perioperative period (Sjoberg et al., 2015).

***2.3 Wider family involvement and or/support*.** Adopting a family-centred approach, having support from the wider family, and discussing treatment options with family members (partners, siblings, grandparents, child) were reported to be facilitators to SDM (Papiez et al., 2021; Waite et al., 2022; Heath et al., 2016). It was further mentioned that it is helpful to formalise the inclusion of families as part of the child's MDT by systematically incorporating patient and family perspectives within MDT case presentations (Heath et al., 2016) and to learn about goals from the child's perspective and include care-givers goals within this (Almoajil et al., 2022). In contrast, SDM was also felt to give 'rise to conflict' In some situations, if one parent was perceived to be more agreeable to surgery, than the other in these situations (Heath et al., 2016).

3. Communication

3.1 Clinician skill in navigating SDM discussions and Interpersonal Characteristics.

The interpersonal qualities of clinicians were noted largely across papers as a facilitator to SDM. Physician empathy, kindness, genuineness, availability, re-assurance and understanding, were all noted as facilitators to engaging in SDM (Atsaidis et al., 2022; Boss et al., 2017; Sjoberg et al., 2015). The importance of clinicians embodying these interpersonal qualities within SDM, are illustrated in the following quotes: "I remember the ENT [physician] had the bedside manner. Understanding, wasn't rushed. That's a lot for me" (Boss et al., 2017, p15), '"It was very welcoming…Everybody was very, very nice. It eased our fears and made us so comfortable." (Boss et al., 2017, p15). The interpersonal nature of the clinician in this instance, was reported as a facilitator, as it was felt to have created a sense of comfort and ease for the patient.

In contrast, clinician apathy to engage in open dialogue, arrogance, or being perceived as being judgemental, were reported as barriers to engaging in shared decision-making (Waite et al., 2022; Boss et al., 2017). This was illustrated by Waite et al., (2022), through the following quotation, "the consultant laughed at me…" (p386) and by Boss et al., (2017) "I felt a little judged by him" (p5) when referring to interactions with clinicians.

Surgeon skill in being able to navigate dilemmas or conflict in therapeutic choices and decisions was considered to be a facilitator to SDM (Loeff & Shakhsheer., 2021) as was the disclosure of the surgeon's level of expertise (Atsaidis et al., 2022). Atsaidis et al., (2022), put forward this argument as follows "it is important to consider the social forces and the power imbalances inherent to the healthcare system, such as differences in role, status and knowledge which can easily undermine the effectiveness of informed consent. With regards to surgical trainees, they must clearly state their role, share their level of training, and discuss with the patient and family that they are training under an attending surgeon." (p842).

3.2 Language and communication. The use of acronyms and medical jargon have been reported to inhibit shared decision-making (Carlisle et al., 2022; Links et al., 2020) and the importance of using layman language, both verbally and in written communication is expressed (Links et al., 2020; Heath et al., 2016). Using a slow conversational pace with frequent repetition of information were also remarked on as being facilitators to shared decision-making (Carlisle et al., 2022). Parents feeling that they had not been listened to, or feeling excluded from discussions about their child's care, were in contrast, reported as barriers to engaging in SDM. Smith et al., (2013c) presents the following quote "They don't seem to take on board what you're saying, that's my feeling. No, they really have their own agenda and that's what we are on now, their agenda" (Smith et al., 2013c, p1308). Timmermans et al. (2018) also references a quote from a psychologist, acknowledging similar feedback from patients, "Some former patients will say, "Mm, you know I would have wanted to be more involved." (p525). The importance of working collaboratively, is highlighted further by Smith et al. (2013c), summarising this with a quote from a Junior Nurse, "I think they should be involved to some degree and you need to listen to them and explain and usually they are on the same page as you anyway." (P1308).

4. Additional sources of support

4.1 Accessing peer information/support and online communities. Having the opportunity to hear from other parents whose child had undergone treatment, accessing information from peer support groups and clinicians making other families available for consultation were found to be facilitators to engaging in SDM (Lecouturier et al., 2015; Timmermans et al., 2018). Waite et al. (2022), present the following quote from a parent in highlighting this, “Having the opportunity for parents to talk to or find out about other kids who have had it, other kids who have had it and it hasn't worked for them or actually something hasn't gone right for them and other parents who've made the decision not to have it and what happened with their child. And then you make a rounded decision, not a decision on, ‘This is the best thing ever, it's good, it's got to be done’.” (p385).

It was further suggested in one paper that educational interventions for patients and families may be beneficial in facilitating decision-making, making reference to patient testimonial videos in particular (Samanta et al., 2022), “It’s difficult for us to understand or empathize or wrap our brains around, but testimonial videos put ourselves in somebody else’s shoes and understand what’s going on. So, I think for the right patient, it might be helpful. (Neurologist)” (p17).

4.2 Mental health, Stress, and psychological needs**.** Included papers also made reference to considering the psychological and emotional experiences of families and or/clients during SDM, with it being raised that clinicians should be mindful and aware of parental stress and anxieties (Timmermans et al., 2018). Offering psychological support to patients was discussed, with it being suggested that this would offer a space to discuss decisions away from medical clinicians (Waite et al., 2022), “Many parents were social media users and joined social networks to seek and share information. These online forums offered parents the potential to have dialogues, in lay language, ‘independent of a medical person’”. (p385). This further emphasises, the role of accessing peer support and the provision of information online, but also the importance of adapting communication styles in enabling SDM.

Discussion

The current review aimed to explore the perceived barriers and facilitators to engaging in SDM when making decisions specifically about paediatric surgery, looking at both the perspectives of patients and families, as well as those reported by clinicians working in the field. In support of previous research, the findings from this review and thematic synthesis identified similar themes centred around the provision of information, having a trusting relationship with professionals and the importance of time and frequency of discussions. Furthermore, language and communication, accessing peer support and being provided with an evidence-base and statistics when making decisions about surgery, were also discussed in relation to barriers and facilitators to engaging in SDM. It is suggested that families experience surgeon recommendations as helpful, with it being important for clinicians to be aware of the psychological and emotional experiences of clients during SDM.

Previous research has also discussed patient related factors that may pose a barrier to engaging in SDM such as the perceived ‘unacceptability’ of asking clinicians questions and believing that it is the clinician’s role to make decisions. Reported clinician related factors have also included, assuming that the patient understands the information being given and being of the opinion that patients or colleagues would not want to engage in SDM.

In support of previous research, the findings from this review suggest that clinicians may perceive parents to have adequate knowledge, which may hinder honest discussions and may prevent parents from considering alternate treatment options (Leu et al., 2021). For example, parents who had a predisposition to choose tonsillectomy for their child, were also found to engage less in the decision-making process and were found to ask less questions in the study. It is suggested that clinicians may support clients during the SDM process, by providing opportunities for families to ask questions about their child’s care and specifically prompting and actively soliciting questions and concerns from parents (Carlisle et al., 2022; Atsaidis et al., 2023; Timmermans et al., 2018). Similarly, in interviewing children about their experiences of the perioperative period, clinicians asking children how they are perceiving the situation and if they had any questions, was also noted as a facilitator to engaging in SDM (Sjoberg et al., 2015). Findings from the current review also suggest that it may be beneficial to provide ‘frequently asked questions’ information to patients (Heath et al., 2016).

In considering how clinicians view parental comprehension and desire to participate in SDM, the current review suggests that surgeons may underestimate parents’ preferences to share decisions, with parents desiring SDM but having little involvement in some cases (Links et al., 2020). Clinicians perceiving parents to have adequate knowledge was also reported to hinder honest discussions between clinicians and parents and was reported to prevent parents considering alternative treatment options (Leu et al., 2021). Although the current study also found that some families prefer surgeons to make decisions (Papiez et al., 2021), it is suggested that it is helpful for clinicians to gauge parental comprehension (Atsaidis et al., 2022), and to tailor decision-making roles to patient’s needs (Links et al., 2020). It was also suggested that surgeons should develop a good understanding of the patient’s social and cultural background in being able to align with the patients’ values, beliefs, and expectations (Loeff & Shakhsheer., 2021).

Since the COVID-19 pandemic, there has been greater utilisation of technology observed within the NHS (Hutchings, 2020) with the NHS long term plan, outlining how services will be redesigned with the aim of avoiding up to a third of face-to-face outpatient visits (NHS England, 2019). In considering the role of technology, the current findings report that families may feel excluded when health professionals discuss their child's care, for example, during ward rounds and care planning (Smith et al., 2013c). It is also reported that parents do not feel involved in initial discussions regarding surgical candidacy and that parental views are often not sought until after the team have discussed treatment options (Heath et al., 2016). The findings from this literature review, suggest that increased opportunities for virtual communication, for example, having a virtual presence during surgical rounds, may enhance SDM (Carlisle et al., 2022). Parents also expressed a preference for being involved in decision-making from the 'out-set,' proposing an early and softer introduction of surgery as a possible treatment option (Heath et al., 2016).

In considering how clinicians engage with online platforms, suggestions were also made regarding bridging the gap between parents and professionals on social media platforms (Waite et al., 2022) and using social media posts to provide support and resources to prospective patients and families (Samanta et al., 2022) which, overall, enhance shared decision-making.

Clinical implications

Adding to the existing evidence base, this review specifically focussed on understanding the reported barriers and facilitators to engaging in SDM in paediatric surgical settings. The findings have implications with respect to how clinicians communicate, share information and connect families with wider support networks. In addition, the results highlight aspects of language and interpersonal skills impacting on SDM and raise the importance of considering the cultural and psychological needs of the family. Paediatric surgical teams may wish to consider SDM interventions and identify further training needs in engaging patients and families in decision-making. Clinicians may also look to consider how to increase involvement from the young person and their support networks within SDM, for example through including the paediatrician within discussions or offering young people and families the opportunity to engage in SDM, which may include virtual wards rounds. Taken altogether, it is hoped that these recommendations will enhance SDM practices and contribute to better outcomes for patients.

Strengths and limitations

To the author’s knowledge, this is the first systematic review of its kind to synthesise the literature on shared decision-making and the reported barriers and facilitators to engaging in shared decision when focussing specifically on paediatric surgical contexts. The findings arising from this review, support the existing literature which has explored barriers and facilitators to engaging in SDM, but contribute more contemporary findings when applied to paediatric surgery. The review benefits from implementing a qualitative design, allowing the author to synthesise rich data from multiple perspectives.

A limitation of the review is that the research was largely conducted in the UK or USA. Owing to the differences in different healthcare systems worldwide, and access to healthcare, funding and resources, the findings are limited with respect to their generalisability. Similarly, when considering the heterogeneity of participants across studies, information relating to ethnicity and socioeconomic status was not collected, and in some studies, respondents were largely female. The study may therefore not represent the experiences of some communities, particularly when considering health inequalities and barriers that may present for some patients and communities, in accessing healthcare, prior to attending for discussions with clinicians around surgery. Furthermore, it is acknowledged that across studies the paediatric sample covers a significant age range. In the study by Carlisle et al (2022) for example, the age of the population being studied ranges from 1 day old, to 13 days old, and in the study by Links and colleagues (Links et al., 2020), the population being studies ranges from 2 years old to 17 years old. Although in the study by Boss and colleagues (Boss et al., 2017), there is mention of surgeons sitting down with young people and colouring whilst discussing surgery, and providing comfort, there is little mention across studies of how SDM processes may be adapted for different age groups and whether there are unique barriers and facilitators that pertain to engaging children and young people in SDM specifically. With the exception of the paper by Sjoberg and colleagues (Sjoberg et al., 2015) which focussed on child perspectives specifically, it is recommended that further research focusses on the barriers and facilitators to engaging children and young people in SDM, supplementing what is already known from parent and healthcare professional accounts.

Conclusion

Discussion and conclusions in relation to the main aim

The perceived barriers and facilitators to engaging in SDM, as reported by clinicians, their clients, and their families, are not mutually exclusive. Patient information needs are highlighted by both clinicians and patients, where the importance of information being timely, clear, specific, and tailored to patients needs is acknowledged. With the rise in technology, clinicians were found to report that patients and their families often seek out their own research. With this in mind, the review highlights the role professionals have in supporting families and young people to engage with online communities and patient groups and to direct families to trustworthy and reliable sources of information in preventing misunderstandings. Connecting families with other patients who have lived experience of going through the decision-making process, was found to be a facilitator to SDM and it is suggested that clinicians may be well positioned to bring families and patients together in sharing their lived experience.

Specific barriers and facilitators, reported by patients and their families, centred around becoming more involved in the decision-making process, for example, through clinicians providing opportunities for families and young people to make decisions from the outset of their care and to have a virtual presence during surgical rounds (Carlisle et al., 2022; Heath et al., 2016). In involving young people and their families in discussions, it is important for the patient's and families' values and beliefs to be considered, which may also include inviting involvement from the wider family and systems around the patient, such as including other professionals e.g. community paediatrician in the SDM process. The importance of allowing adequate time and the frequency of discussions, is also reported by patients and their families, in which it is reported to be beneficial to the SDM process to repeat information, check understanding, and offer repeat visits to address patient queries and concerns.

In conclusion, the findings from this systematic review, support the recommendations outlined in NICE guidelines (NICE, 2021) for enhancing SDM. Recommendations for intervention include offering pre-consultation interventions, Interventions to improve health literacy, preference/value elicitation, third person support (involving other professionals/family members in SDM), patient activation (referring to a patients self-assessment of their understanding, knowledge and confidence in being able to manage their own health) and documentary intervention (collection of ongoing data, that is fed back into SDM, for example, considering findings from trialling different treatments or interventions). It is recommended that future research evaluates the effectiveness and impact of offering these interventions on enhancing SDM, which may focus on paediatric surgical populations.

Chapter Three: Bridging chapter

Chapter two presents the findings from a systematic review, which explored the reported barriers and facilitators to engaging in shared decision-making (SDM) for paediatric surgery as reported by clinicians, children and young people and their families. The current chapter will discuss the relevance of these findings, applied to cleft lip and palate services (CL/P) and decision-making for orthognathic surgery (OS).

Four analytic themes and 11 sub-themes were identified through the process of conducting a thematic synthesis. Analytic themes included being aware of the patient's information needs, enabling effecting communication, promoting access to support and being aware of the patients networks and support systems. In discussing these themes further, the importance of involving patients and their families in SDM and all parties feeling listened to and heard is stressed (Sjoberg et al., 2015). It is also raised that clinicians should consider the patients values, beliefs and cultural background when facilitating SDM (Waite et al., 2022; Loeff & Shakhsheer., 2021; Smith et al., 2013c).

Consistent with the themes identified in the systematic review, research exploring the involvement of young people with CL/P n decision-making for elective surgery, highlights the importance of hearing the young person's voice and being mindful of external pressures impacting on young people during SDM (Bemmels et al., 2013; Kapp-Simon et al., 2015). Clinicians and parents were found to be influential during SDM of which young people reported to experience pressure from parents and feeling 'left in the dark' by professionals (Wogden et al., 2019). Furthermore, young people with CL/P have reported that they just "went along" with the decision for OS and perceived the discussion with clinicians about OS as a 'recommendation for surgery,' which was normalised as 'routine' and part of the treatment pathway by professionals (Acum et al., 2018). These findings therefore bring awareness to external pressures influencing SDM and raise questions as to how clinicians consider these dynamics, when supporting young people in their decision-making for OS.

In further considering the influence of support networks during SDM, the results of the thematic synthesis also highlight the importance of being aware of the psychological wellbeing of patients and families; and being aware that families may seek out information online (Loeff & Shakhsheer., 2021; Lecouturier et al., 2015; Timmermans et al., 2018). With it being acknowledged that families and patients will regularly seek out information online, the importance of accessing reliable information is noted (Timmermans et al., 2018). Conversely, receiving information that is felt to be in contrast to the advice provided by clinicians, was reported to be a barrier to engaging in SDM (Lerret et al., 2016). It is therefore important to consider the role clinicians may have, in signposting and providing opportunities for patients to connect with online communities, as well as acknowledging the impact this may have on SDM for OS.

In reviewing the literature surrounding CL/P and psychosocial factors, health inequalities impacting on access to CL/P services are also raised (Abbott et al., 2011; Zaluzec et al., 2019; Smillie et al., 2015). These include the availability of services within different areas, transport and childcare considerations, and consideration of language, literacy, stigma, and previous experiences of accessing services. Different cultural beliefs around surgery, may also present, including viewing CL/P as being a ‘gift from god’ and spiritual interventions and traditional medication being sought (Hasanuddin et al., 2023). It is therefore of importance, to understand how these factors may be considered by clinicians working with this client group, with a particular understanding of these factors and how they may present at the transition period when supporting young people in decision-making for OS.

In addressing the discussion around ethics applied to CL/P and OS raised in chapter one, as young people experience a shift in decision-making and enhanced responsibility, it is also important to explore how the balance may be maintained between autonomy, beneficence and capacity within SDM discussions at the transition period (Strauss, 2002). It is acknowledged that parents and or/caregivers have a significant role during SDM in the earlier stages of the pathway, however it is also of importance to explore, how these ethical dilemmas may be considered and thought about at the transition period whereby there is often delegation of responsibility to the young person.

Acknowledging the significant shift in decision-making roles that occurs during adolescence, the current study focusses on the transition period in particular and clinicians' consideration of psychosocial and cultural factors. The current study also explores barriers and facilitators that may arise when supporting young people around decision-making for orthognathic surgery and associated ethical dilemmas that may present during the decision-making process.

Chapter Four: Empirical Paper

Empirical paper: Decision-making at the transition period: Widening the lens around psychosocial and cultural factors in cleft orthognathic surgery.

Abstract word count (Including key words): 259

Paper word count excluding tables: 7,998

Prepared in accordance with the requirements for submission to The Cleft Palate-Craniofacial Journal (see guidelines in Appendix 9)

Abstract

Objective: To explore what factors are considered by MDT professionals working in Cleft Lip and Palate services (CL/P) when supporting young people in their decision-making for orthognathic surgery (OS) and to explore how professionals understand and manage ethical dilemmas that arise.

Design: A qualitative design was employed in which semi-structured interviews were conducted remotely with NHS staff working in UK cleft services.

Participants/Setting: Nine participants were interviewed across four NHS cleft services. The sample consisted of three Speech and Language Therapists, three Orthodontists, and three Surgeons involved in decision-making for OS.

Results: Thematic Analysis (TA) was undertaken to adhere to guidance by Braun and Clarke (2006). The researcher aligned with interpretive-constructive approaches, illuminating the experiences of clinicians in relation to surgery decision-making and consideration of psychosocial factors. TA revealed three key themes and seven sub-themes, including ‘navigating the decision-making process,’ ‘Team-Centric’, and ‘Health Inequalities impacting on access to cleft services’.

Conclusions: The findings highlight the complex nature of balancing patient autonomy and beneficence during the decision-making process for OS whilst bringing awareness to factors that may pose a barrier to decision-making such as the presence of health inequalities, power dynamics and conditions of the MDT environment. The importance of optimising the shared decision-making environment and creating opportunities for liaison, particularly earlier on in the pathway, are discussed. It is recommended that future research explores the impact of health inequalities on access to cleft care in greater detail, recognising those that are currently disadvantaged in their treatment journey.

Key words: Cleft Lip and or/Palate, decision-making, Orthognathic Surgery, Young People

Introduction

A cleft lip and/or palate (CL/P) is one of the most common craniofacial conditions, reported to affect around 1 in 700 births per year (Yilmaz et al., 2019). In the UK, patients born with a CL/P are commenced on a 20-year treatment pathway in which early surgery is required at around 3-6 months old to close the gap.

At the transition period (defined here as a period of significant change and adjustment, referring to young people aged 16+ whereby surgery decision-making is delegated by a parent and/or carer), young people may wish to elect for jaw surgery (also known as orthognathic surgery, 'OS'). This is a procedure, usually performed once the young person has finished growing which involves re-aligning the jaws and is usually offered to patients experiencing adverse functional or aesthetic consequences.

The process of decision-making for OS, requires surgeons discussing the advantages and risks to undergoing surgery and ensuring that the patient has a good understanding of what surgery may entail. OS has been associated with aesthetic and functional improvements, such as achieving greater facial symmetry and jaw alignment whilst it is also reported that OS may lead to improvements in speech, breathing, quality of life and psychosocial functioning (Ganoo and Sjöström, 2019; Pellby and Bengtsson, 2024; Cremona et al., 2022). Whilst post-operative outcomes are largely positively reported (Zamboni et al., 2019), risks to undergoing OS can include injury to salivary ducts, post-operative infection, blood loss, scarring, nerve injury, deterioration in speech and relapse (Khechoyan, 2013; Ferri et al., 2019; Dalston, 1984; Zaroni et al., 2024).

It is also important to consider the impact on education, employment and the social lives of young people in making a decision to undergo surgery. In interviewing young people about their experiences of decision-making, young people raised how this required time out of work and impacted on them being able to partake in activities such as sports. Young people also shared that they considered taking a gap year to accommodate appointments and treatment (Acum, 2018). Careful planning with consideration of these factors, is therefore required, requiring a multidisciplinary (MDT) approach.

Due to the prolonged nature of the treatment pathway, often involving multiple surgeries, treatments, and interventions; young people will be used to their parent and/or caregiver, making healthcare decisions on their behalf. The transition period is often the first time a young person is delegated responsibility for decision-making, of which the gravity and commitment associated with making a decision for OS is acknowledged (Acum, 2018). In acknowledging the commitment of young people to the treatment pathway, it is paramount for clinicians to have insight into the young person's internal and external world, and to understand their motivations and expectations for surgery. This may be influenced to a greater of lesser extent, by social, cultural, and psychological factors, impacting on the shared decision-making (SDM) process. Whilst the need for OS is not limited to young people who have a CL/P, this group of young people are of interest, due to the longevity of treatments across their lifespan and with OS being offered at a point of transition whereby responsibility for surgery decision-making is often delegated to the young person. This empirical study aimed to explore clinicians consideration of psychosocial and cultural factors and associated ethical dilemmas that arise, when supporting young people in their decision making for OS.

Psychosocial factors and adjustment to Cleft

Social stigma and psychosocial effects associated with CL/P are reported amongst adults and children, however the literature is reported to be largely inconclusive (Stock and Feragen, 2016; Stiernman et al., 2019). Some of the reported psychosocial effects associated with having a CL/P include experiencing difficulties with self-concept, attachment, interpersonal relationships and experiencing mental health difficulties (Hunt et al., 2005). In consulting with patients with CL/P and their families, patients reported to have experienced difficulties with self-confidence, initiating conversations and teasing. Amongst patients aged between 15 and 20, 73% reported their self-confidence to have been affected as result of experiencing a cleft, which rose to 91% amongst 20-year-olds (Turner et al., 1997).

In interviewing parents and clinicians about their views on psychosocial and educational outcomes for their child with CL/P, clinicians expressed concerns around how the young person might cope with ‘”difference,” low self-esteem and acceptance of themselves. Previous research has reported that clinicians wish to address psychosocial factors but feel restricted in doing so due to time limitations and feeling that they did not have the right environment, knowledge, or education to do so (Clarke and Cooper, 2001; Rumsey and Harcourt., 2004). Consistent with these reported barriers, is that none of the clinicians in the study were reported to have received formal training regarding psychological factors or adjustment to cleft, indicating further training may be beneficial (Stiernman et al., 2019).

In exploring how Consultant Orthodontists view psychological support for orthognathic patients, 90% reported to feel that ‘some’ of their patients would benefit from a referral to psychology, with 41% referring approximately 10% of their patients for support.

Similar to the findings from Stiernman et al (2019), 81% of Consultant Orthodontists reported to have no teaching or training in psychological assessment and management (Juggins et al., 2006) highlighting a need for further training.

Ethical dilemmas associated with surgery decision-making.

SDM is an essential and ethical obligation of Craniofacial Surgeons and extended members of the MDT when supporting patients in their decision-making for OS. Involving patients in the decision-making process not only ensures that surgeons are operating in cases where clients are invested in the procedure and recovery process, but this avoids operating in cases where the patient is satisfied with their appearance and is disinterested in surgery (Bennett et al., 2022). The Montgomery ruling outlines how patient’s should be supported to make decisions based on their values and preferences, being told what they “want to know” as opposed to what the doctor “thinks they should be told” however it is acknowledged that this poses a difficult balance for clinicians in promoting patient autonomy whilst acting in the patients best interest (Chan et al., 2017).

Other ethical dilemmas clinicians may face include the patient, family and clinician holding different views around treatment and parental autonomy acting as a barrier to SDM (Bemmels et al., 2013). Furthermore, in a study looking at young people’s decision-making experiences around end of pathway cleft surgery, the researcher noted that once a malocclusion was pointed out, the young person questioned this aspect of their appearance (Acum, 2018) suggesting that there may be ethical considerations around informed consent and the decision-making process.

Objectives

The current study aimed to explore what factors are considered by different MDT professionals working in CL/P services when supporting young people in making decisions around OS, whilst also exploring how different professionals understand and manage ethical dilemmas that may arise during these interactions. The current study addressed the following research questions:

- How do clinicians from different backgrounds experience and manage ethical dilemmas when supporting young people in making decisions about elective OS?
- What factors do clinicians consider when consulting with young people about OS?
- How does clinician awareness of psychosocial factors impact on the decision-making process when supporting young people in decision-making for OS?

Ethical Approval

Approval was sought from the Faculty of Medicine and Health Sciences REC at the University of East Anglia (Appendix 10) and Health Research Authority (Appendix 11). Local approvals were sought from research and development at the participating NHS sites.

Design

A qualitative design was employed in which semi-structured interviews were conducted with Orthodontists, Speech and Language Therapists (SLT's) and Orthognathic surgeons involved in the decision-making process for OS. A qualitative design was employed in allowing for in-depth exploration of clinician perspectives. An interview topic guide (Appendix 12) was developed following consultation with a Clinical Psychologist, Orthodontist, Surgeon and SLT working in the field and was further role-played in ensuring that this flowed accordingly and felt appropriate for the setting.

Sampling and inclusion/exclusion criteria

Three Surgeons, three Orthodontists and three SLT's were recruited across cleft services at four of the five participating NHS sites. This sample size was decided upon in accordance with recent guidelines provided by Braun and Clarke (2013, p.50) which suggests recruiting 6-10 participants for small projects involving interviews. In considering the limited availability of staff working in cleft MDT's and number of professionals in post, a target of three clinicians from each professionals group was set. Clinicians were eligible to take part if they had been involved in the decision-making process for OS, had access to Microsoft Teams and consented to being audio and video recorded. Clinicians were not able to take part if they did not work for one of the participating sites, did not consent to being recorded or if they did not have access to using Microsoft Teams. A template email (Appendix 13) and Participant Information Sheet (Appendix 14) was disseminated to clinicians in each team by the Principal Investigator. Following this, clinicians made contact with the Chief Investigator (HC) directly in expressing an interest in taking part in which they were followed up and consented into the study.

Interviews

All interviews were conducted remotely by the Chief Investigator (HC) using Microsoft Teams. Interviews took place from August to December 2023. A semi-structured topic guide was implemented, and questions were framed around communication and team working, perceived barriers and facilitators to engaging in SDM, professionals understanding and consideration of psychological, social, and cultural factors during the decision-making process and understanding and management of ethical dilemmas (see Appendix 12).

Participants signed a consent form prior to the beginning of the interview, in which they consented to the interviews being audio and video recorded and were informed how their data would be used and stored. Participants were assigned a Participant ID and information was anonymised during transcription in maintaining confidentiality.

Participants

Nine clinicians were recruited across four NHS cleft services. The sample consisted of three SLT's, three Surgeons and three Orthodontists due to their significant involvement in decision-making at the transition period. Clinicians reported experience working in cleft services, ranged from three years to 20+ years. A third of the participants were male.

Analysis

Three major themes and seven subthemes were identified through the process of conducting a Thematic Analysis (TA), adhering to the protocol outlined by Braun and Clarke (2006). The researcher (HC) first listened back to each interview in familiarising themselves with the data, recording their Initial thoughts and reflections during the process. Following this, the researcher started line by line coding, identifying initial themes arising from the data. A coding framework was developed in defining themes and these were revised as an ongoing process (Appendix 15). During the course of defining and revising themes, themes were brought to supervision and discussed. A table representing the final themes is provided below.

Results

Table 4: Themes and subthemes

Theme	**Subthemes**
Theme 1: Navigating the decision-making process	• *Finding a 'balance'* • *External factors impacting on the decision making process.* • *Managing positions of power* • *The process of eliciting motivations for surgery.*
Theme 2: 'Team-centric'	• *Opportunities to foster team communication.* • *Valued contribution of MDT members*
Theme 3: Health Inequalities impacting on access to cleft care.	• *Equity and barriers to accessing care*

1. Navigating the decision-making process

Across interviews, it became apparent that many aspects of the decision-making process elicited feelings of discomfort for clinicians. Clinicians reported to experience 'internal conflict' when balancing autonomy and beneficence, or when patients presented to clinic having been 'told' OS was routine. Clinicians expressed the importance of understanding the young persons motivations for surgery and voiced discomfort in being asked to operate when these reasons were not clear.

In engaging young people in SDM and eliciting motivations for surgery, clinicians were aware that young people may not 'readily' volunteer their reasons for wanting to proceed. Clinicians spoke of exploring concerns that may be 'unvoiced' such as those related to aesthetic or cosmetic reasons. Clinicians were also mindful that the presence of lots of professionals in one room, and layout of the room (e.g. positioning of the dental chair) could create power dynamics and pose a barrier to SDM.

1.1. Sub-theme- Finding a 'balance': As clinicians spoke about the transition period, acknowledging the delegation of responsibility from parent and/or carers to the young person being positioned as decision maker, themes emerged around balancing patient autonomy and acting in the patient's best interest (beneficence). Clinicians acknowledged a shift in paternalism but were mindful of their role and responsibility in making the 'final decision.'

> I'm constantly challenged, because of the nature of our work. It's a constant challenge to try not to be to be paternalistic about things, but then there are some times when you just have to say no and so finding the right balance. (Participant 7).

This was further expanded on in the following quote, in which one clinician referred to the decision-making process as deciding between 'can' vs 'should'.

> Can you do surgery? Yes, but should you do surgery? And I think that's always important, no matter what type of surgery you're doing. (Participant 5).

Clinicians also shared their concerns around not wanting to make a situation 'worse' and worrying if they had done the 'right thing', of which the desire for 'certainty' was also noted.

> It's not my own ego, but it is hard because you're like, have I done the right thing operating on her and I, you know, she has good results. (Participant 5)

> What are the potential complications if I do something that leaves them worse off for something that isn't, you know, required to save their life or then... then that's…that's the responsibility that I have, or our team have. (Participant 7)

> I'm probably more risk averse than many of my younger colleagues would be, but I would rather wait until a patient is able to clearly say yes or no, rather than you know, going ahead and say, well, let's just try it, lets just see how you go (Participant 3).

On the theme of 'finding a balance' clinicians also shared their experiences of supporting patients from different cultural and religious backgrounds. Below, a clinician shares their experience of working with a patient who identified as a Jehovah's Witness, of which the clinician accessed an advocate in supporting them and the patient to understand each others' views around the risks involved in proceeding with OS.

> We had a Jehovah's Witness advocate and that was very helpful in terms of helping me understand why it was important as well, so... because it's very easy to say, well, this is my belief and...I don't understand why you can do this to shift to.... It's not about my belief, although you do have to acknowledge that your actions could impact on me...so there we did have a bit of a conversation about, but what does this mean to me? What does this mean to me If you were to die on the table? (Participant 2)

In sharing experiences of supporting families from different cultural backgrounds, a clinician made reference to families who position clinicians as 'expert'. In discussing working with a family from a Roma background, the following example was shared:

> There are some groups that still regard sort of anything a health professional says as they are the health professional It's not my place to challenge it, I actively encourage them to challenge it, but it doesn't sit comfortably with some groups. (Participant 3).

Clinicians also expressed discomfort when young people presented to clinic having been told they 'need' OS or that this is just the 'next step' in the treatment pathway.

> often you hear well, I've just been told I need surgery and I don't want to operate on someone who's only having this done because they've been told they need it. So, it's really important to me to kind of see what bothers the patients, what their concerns are and see if surgery is going to help with those. (Participant 5)

The presentation of co-morbid mental health difficulties, and who holds responsibility for supporting patients with their mental health, was also highlighted as an ethical issue with respect to 'maintaining a balance' and keeping people safe.

> Severe depression, which is triggered by cleft that's still severe depression that needs treating in mental health services not in cleft services. So sometimes there's a bit around ...boundaries... there that... where do we sit and that and how do we keep patients safe? (Participant 2).

In discussing body-dysmorphia, the clinician raised how it may be difficult to ascertain whether surgery will achieve the desired outcomes of the patient, raising an ethical dilemma.

> You've got to, you know, are you dealing with the body dysmorphia...? Probably. And it's harder to say that when you're doing cleft because there is actually a physical deformity... that you have congenital deformity…but you do still get patients that

> have altered perception and if they have altered perception...is doing surgery actually going to give them a result that they will be happy with? (Participant 2).

1.2. Sub-theme - External influences impacting on decision-making: As families and young people increasingly seek out health information online, the pros and cons to accessing information online is debated. Clinicians acknowledged that online communities and social media platforms may create a space for patients to come together, share narratives and ask questions, however concerns around the regulation of online platforms were raised. Clinicians were also aware of the possibility of misinformation being shared online, and noted the impact that social media platforms can have on young people's sense of self and appearance.

Social media platforms such as TikTok and Instagram were reported to influence young people's perspectives on their appearance, bringing awareness to asymmetry and desire for surgery for aesthetic reasons.

> Selfies aren't too bad, but it's more when people take photographs of you, that's when you see the asymmetry, because it's like you're looking at another person, not a mirror image. And your asymmetry... if you think about it... that's your midline. If you're over there. Yeah. So, it's only that much difference in a photograph. You're over there. So, it's actually that much different from what they're used to seeing. So, photographs are double. They magnify the difference. (Participant 2).

> I wonder whether that's just, you know, the Instagram generation, if you like, the people who are much more focused on faces than perhaps they ever have been. Boys, girls alike, you know, they often do want to go through treatments to average out their face to balance out their face. (Participant 6).

Concerns were also raised around online platforms being unmoderated, in which patients may be subject to misinformation, or may use online platforms to express dissatisfaction with their care or experiences with services and professionals which may adversely influence other patients and create feelings of discomfort for professionals.

> Standard concerns about things being unmoderated, advice that we wouldn't necessarily recommend is being shared quite readily without anybody there to say, oh no, no, don't do that. (Participant 8)

> And so, they just kind of offloaded in this group and its sort of hard to read because you can't then as a medical professional contact that parent and say, ohh, I heard you were unhappy about this. Sorry about that, because there has to be those boundaries of if they want to speak to us, to complain about it, that's a different matter. (Participant 8).

In being aware of this, clinicians spoke of signposting patients to the British Orthodontic Society and patient experience videos in accessing reliable and up to date information.

> We do try to stop patients accessing. You know non sanitized, you know, Internet publications of whatever sort. We try to say, listen, if you're gonna read anything online, make sure it's sort of British stuff. Orthodontic society cleft world stuff so that your as sure as you can be that you're getting what we would be considered as our treatment modalities rather than what might be happening overseas or somewhere else and whatnot. (Participant 6)

Although concerns were raised with respect to seeking out health information online, patient communities and social media platforms were also noted as being a helpful for patients in terms of hearing peer experiences of undergoing OS.

> But I think there's a there's a sense of it's good to have some other agency or some other group that speak to the patients or inform the patients about that which is useful in terms of they're getting a view from some other forum. I know one or two patients who've done who've got their information from TikTok. You know, they followed someone who is, who has shown their experiences on TikTok, of having had the surgery done. (Participant 4).

1.3. Sub-theme -The process of eliciting motivations for surgery: Psychological safety is highlighted as a central component in patients feeling able to disclose their concerns during SDM, of which clinicians were aware that understanding the young persons concerns, many involve 'picking out' or 'teasing out' motivations for surgery.

> Under all that, there are reasons why people probably would like to have the benefits of having jaw surgery. But I think it's really important to kind of pick that out. You always want somebody…I mean, that's just how it was trained. You always wanted to get it out of the patient. Why they... why they want surgery. (Participant 5).

Another clinician also describes this process as 'deep diving' into the 'layers' of concern.

> Really deep diving into…the burden that these patients carry and the sort of layers of not dissatisfaction but the layers of concern related to appearance and how they're perceived and how many of those are things that we can improve and how those are

> related to things that we look at and go, I could do you the best bite and the best jaw surgery, but I sense you're still not going to be entirely happy with things, and it's about sort of riding out those decisions. (Participant 3).

One of the reasons clinicians felt patients may be more hesitant to share their concerns, is anticipating that patients feel it is not acceptable to proceed with surgery for aesthetic or cosmetic reasons, with gender differences also being acknowledged with regards to pragmatism, awareness of emotions and expectations around surgery.

> There is a perception within the NHS that you can't really ask for things for an aesthetic basis. It's all got to be functionally related…. (Participant 3).

> You know, it's a cosmetic thing, why should I have access to cosmetic treatment?' (Participant 2)

> they've boxed it because as a bloke they shouldn't be going down that pathway of caring what their nose looks like, of being vain, whereas the girls handle it a lot more. They're just much more with it. They're just more. Yeah. Don't like my nose, I want something done about it. I have the right to want something done about it. Why shouldn't I have a good nose? You know., so they're... in some ways... they are... are much more emotionally in-tune and pragmatic about what they want than the boys are, the boy's pragmatism boxes them in. (Participant 2).

Asking open questions, and the importance of using language that does not imply positionality towards surgery, was highlighted as an important factor in supporting young people to make decisions and avoiding creating feelings of difference or 'dissatisfaction' with appearance.

> You could have someone who's quite happy and then you tell them... that they're not right. So ...you're creating a problem and... that that is challenging. How do you? What language do you use? rather than saying right/wrong, you might say ‘your top teeth are behind your bottom teeth. Are you happy with that appearance? Can you function OK,’ not, it is wrong, and moving away from that concept of they as a patient are wrong in any way or not normal (Participant 2).

Concerns raised around not creating feelings of difference, were also highlighted in the following quote.

> I'm really cognizant of not creating…that awareness that, oh, maybe I look different. Is there something wrong with me? So, I can distinctly remember seeing a young girl, you know… in…and she had no issues with how she looked. She had brilliant confidence, et cetera, and I'm very cognizant that I don't want to create something in her mind. (Participant 5)

In exploring the ‘layers’ of concern, and ‘unpicking’ patients understanding and motivations for surgery, clinicians also spoke about the role of psychology within the MDT.

> but having the psychologist on board has been utterly invaluable because they've been able to sort of dissect out where the areas of concern are rather than kind of, I'm unhappy with everything they've been able to dissect that... well… what elements that are specifically related to your cleft are you unhappy with? what elements like to talk about with the surgeons? What elements are you worried about with your speaking? What elements do you not like about your teeth? and they can break it down. And so, the patient comes back sort of prepared with more specific questions (Participant 3).

The role of psychologists was also raised with respect to supporting clients with autism or learning needs and understanding capacity.

> so I think that is tricky and then you know you every now and again you do you get a patient with special needs, with perhaps ASD or something like that and then picking apart their cognition of what they're letting themselves in for understanding their capacity for decision-making that I think that that is that is a real challenge but that's a really small subset of our patient group. Certainly, those guys I think we would ask the psychologist to help us really with those decision-making process. (Participant 6).

1.4. Sub-theme -Managing positions of power: Whilst trying to elicit motivations for surgery and engage young people in SDM, clinicians reflected on both the advantages and potential drawbacks of coming together with other professionals and the patient in one room. Professionals acknowledged how meeting as a team with the patient provided helpful opportunities to have discussions with colleagues and for patients to observe that the decision-making process requires significant thought and care. In contrast, professionals reflected on how the presence of lots of professionals in one room may feel 'intimidating' and 'overwhelming' for patients, resulting in them being less likely to disclose motivations for surgery. It was also raised that the 'modern day MDT experience' may bring back memories for clients of earlier challenging times in their childhood.

> we do get returning adults who perhaps had a challenging experience as kids and they find that perhaps the MDT environment is a bit overwhelming despite the fact actually that they may... they may see the psychologist beforehand and they prep them for what a…modern day MDT experience is like, but still they say, listen, I can't

> cope with that, that's.. that brings me back to my childhood, which was, which was challenging (Participant 6).

> If you have, you know, like 3 consultant surgeons and a couple of trainees and a consultant orthodontist like, you're not going to bare your soul and be like, oh, actually, I'm really subconscious about this You'd just be like, I'm here to talk about surgery, so I think taking the patient out of that environment and having a you know, less intimidating chat is really valuable, I think. (Participant 5)

The presence and positioning of the dental chair was also highlighted in relation to creating power dynamics between professionals and the patient, of which clinicians spoke about the particular placement of the chair behind a wall and importance of having this positioned at eye level with patients.

> So those clinics do happen with them sitting down and everyone else towering above, and I wonder if that can have a negative impact or a positive one, not you know, the sort of feeling lower down than everyone because they're sat down, but the fact that everything is in the same room and everyone's hearing the same thing. (Participant 9)

> and a dental chair which is behind the kind of half high wall so the patients don't walk on and see a dental chair, they come in and they seated area and we sit them down and we… we make sure it's that sort of...subtle things, but we make sure that we're always at eye level with them, so that the seats that the patient sit on are slightly higher than ours. So that when we are looking, we're on an equal level and we space it to make it sort of as informal as possible. (Participant 3)

Whilst the drawbacks of the MDT environment are acknowledged, conversely, the MDT environment was felt to provide opportunities for discussion and for everyone to hear the same thing.

> We have everyone in the room at the same time. Now the advantage of that is we can have around the table discussions where we can thrash out all these things. (Participant 7)

> I think it's useful for them to see that we have really thought about it, and we've had a proper a discussion about it on the other side of things. (Participant 7).

Whilst holding both the advantages and challenges to meeting as an MDT in mind, clinicians were mindful of the need to create psychological safety.

> It's gonna be about psychological safety isn't it, if they don't know you, why? Why they're going to discuss it? They don't know how it's gonna land. And they've made... sometimes they'll have made assumptions. (Participant 2).

The following quote illustrates the importance of patients having a safe space and creating psychological safety, in being able to understand the information that is given and feeling able to ask questions.

> And you know, the patients came back and said to [clinician] why are they feeding me wafers... if I'm going to be asleep during an operation and she'll say no, it's not like a pink wafer. What they're talking about is a bit of plastic that fits between your teeth. And I've gone. Oh, and the fact that they haven't felt sort of comfortable enough to ask that question reveals quite a lot about how the conversation goes, and

> you know, there is so much that we probably think we're doing right, but without asking the patients, we don't know. (Participant 3).

Theme 2: “Team-centric’*:* Clinicians reflected on the nature of having centralised and bespoke teams, speaking to the implementation of recommendations raised in the CSAG report. Opportunities for liaison with colleagues differed between teams and was influenced by team-set up. Some clinicians felt they had many opportunities to liaise with their colleagues, speaking of the advantages of being able to have more ‘ad-hoc’ discussions. Other teams in contrast, desired more opportunities for team liaison and reported that there were not many opportunities to liaise, resulting in delays to updating patient facing information and discussing feedback. Within these discussions, equity across services, and awareness of health inequalities and barriers to accessing cleft care, were noted.

2.1.Sub-theme- Opportunities to foster team communication: Clinicians commented on the advantages of being able to have ‘ad-hoc’ conversations and liaise with their colleagues, whereas other teams relied more heavily on the support of community-based colleagues and connecting remotely.

> the key to a strong and functional team is that you do have the opportunities just to have corridor conversations and that you do bump into your colleagues. (Participant 6)

> Very dispersed team in that we rely really heavily on the support of our community-based colleagues. We've never had the funding all centralised, so we have very good links across primary and secondary care and that’s really, really important in terms of the way lots of things we do are run (Participant 1).

Other clinicians felt there could be more opportunities for liaison in-between clinics, emphasising the importance of having opportunities to meet together as a team.

> You know, we don't really and actually it would probably be good if we did, because there are a few things that we need to think about and so like, we have a leaflet that we think needs updating and that's almost a year and nothing has happened and probably because we don't have any platform for meeting up in between those clinics. (Participant 9)

> I should make more time to try and set up a way of discussing people… patients... difficult patients with other colleagues because it means… because of the very specialized nature of it…you have to go out of region to find someone who really knows what you're talking about…so that's ..that's one of the things that that I would like to develop (Participant 7)

2.2. Sub-theme-Valued contribution of MDT members: Clinicians highlighted the significance of building relationships with patients across the treatment pathway, of which the role of the Orthodontist was particularly highlighted, due to the level of interaction they may have with families and young people.

> I think, being slightly biased, the orthodontist having potentially known the child... or the patient rather, since they were a child, often may have… quite an insight into that patients…life, their thoughts their, you know their personality and so sometimes the orthodontist can ..can take a bit more of a lead (Participant 6).

The shift from paternalism is also noted within teams, of which a clinician describes the nature of the MDT as ‘everybody is a clinic lead.’

> I'd very much hate to work in a team where, for example, the surgeon led everything and was, you know, was seen as the clinical lead when everybody's a clinical lead because everybody's part to the service and to the input to the care is important. (Participant 6).

Although the value of each speciality within the MDT is noted, themes arose around how SLT's view their role, and how the role of SL's are viewed, in relation to the surgery decision-making process.

> I think we have quite an important role to play in, in, in different ways. So, I think, I think we have an important role to play clinically, in terms of our specialism and in terms of advising. (Participant 1).

> if it was to go in order of a contribution and impact on that patient's journey, then I think probably we would maybe come bottom of the pile. I think possibly, and I think feel like perhaps our psychology team and orthodontists have even more involvement with those patients than we do. (Participant 6)

> Speech and language therapy colleagues tend to do the assessment and just feed in and give some general advice around risk to speech to the patients and… and that's about it. They don't have a huge impact otherwise... and not very much direct influence on treatment planning or what we do with the surgery, I suppose. (Participant 4).

Theme 3: The impact of Health Inequalities on access to cleft care. Whilst reflecting on the nature of centralised and be-spoke teams, clinicians were mindful of the various barriers patients may face in receiving care. Clinicians raised issues relating to equity across services, financial and travel implications, and the provision of services in different regions.

3.1. Sub-theme-Equity and barriers to accessing support: In speaking to health equalities perceived to impact on access to services and cleft care, a clinician highlighted the requirements placed on patients, precluding attending clinic. In particular, the clinician highlights the expectation that young people will be able to access a dentist regularly in promoting ‘good oral health,’ which may impact on being able to attend clinic.

> I don’t see the same mixture of backgrounds in our cleft orthognathic than I would expect from kind of. It’s in the general population now. That may be because there are so many other factors that preclude them actually turning up to clinic. You know, you think of all the hurdles that we put in place. They’ve got to be able to attend appointments, they’ve got to be able to have… they’ve got to be able to see a dentist on a regular basis to have good oral health. They’ve got to be able to engage with the communication from the hospital from the admin team, inviting them to appointments, and I would say all those things…exclude quite a significant proportion of people that we look after (Participant 3).

Consideration is also given to childcare, financial and employment implications that may impact on being able to access healthcare, highlighting the various barriers that may exist, prior to surgery decision-making.

> And we select patients to be seen there by post code and so many of them have said there's no way I’d have got up to the (clinic location). You know, I can't do it with picking up the other kids or getting there or even the bus, even though transport costs are reimbursed on the day it's actually having… It's down to having that

> amount of disposable income that you can pay out bus fares for a parent and child. (Participant 3)

> maybe that's through lack of, you know, opportunity to access our care. You know, I think probably families who are in those situations that, their need to be at work, for example, and they're their ability to get away from work, to bring their kid to treatment. It is definitely much more of a challenge for some than others. (Participant 6).

This was also discussed in the context of the transition period, and how receiving treatment may impact on education, employment in addition to impacting financially on the young person if being required to travel as a prerequisite to having OS.

> When you're a teenager, often they look at it as, oh, I'm going to miss a session of school every six weeks. That's not such a bad thing, but when you are putting yourself under pressure for A Levels or going to university or starting an apprenticeship, and you're looking at whether your employer is going to let you out of work for such regular times, or whether it's going to… leave or how you're going to get there independently and the cost of travel, there are lots of different factors on somebody's desire to do all of that as a pre requirement to the jaw surgery. (Participant 3).

In hearing the narratives from clinicians who work across different regions, it was acknowledged that processes may also vary between sites, which may include the advice and guidance provided and availability of professionals in clinics. In reflecting on equity across services, clinicians raised the following points:

> The other is a lack of consistency in approach across the whole of the [region]. So, there might be inequity in terms of that service because they may not hear the same thing from my local, you know like from my colleagues who do the clinic in (different locations). (Participant 4).
>
> There's a bit of a mixed economy in that regard. Some the clinics I will go to or my colleague who also does orthognathic surgery, goes to and then there will be other clinics that there is no maxillofacial orthognathic surgeon attending. So those patients would most likely have to travel to [other site location] to be seen by the surgeons to get the advice about what they think is possible. (Participant 7).

Discussion

Key findings

The findings offer insight into the experiences of clinicians when supporting young people in their decision-making for OS, of which three main themes and seven sub-themes were identified through conducting TA. Themes included, ‘navigating the decision-making process’, ‘team-centric’ and ‘Health inequalities impacting on access to cleft care’.

Discussion in relation to the main aims

TA revealed a shared experience of ‘internal conflict’, particularly arising from surgeons when being positioned as the final decision maker. Clinicians described the challenging nature of balancing patient autonomy and beneficence, expressing concern around ‘doing the right thing.’ This was underpinned by a desire for certainty in the decision being made, of which clinicians experienced discomfort when positioned as ‘expert’ or when young people presented for OS having been ‘told’ they need surgery. This is consistent with the shift in paternalism to SDM and the Montgomery ruling that ‘patients should be told what they want to know, not what the doctor thinks they should be told’ (Chan et al., 2017). In contrast, this raises a particular challenge for clinicians, when trying to maintain a balance between autonomy and beneficence, exacerbating feelings of discomfort and concerns around ‘doing the right thing’ when positioned as final decision maker.

Previous research exploring the decision-making experiences of young people, suggests they feel ‘tied’ to the treatment pathway, therefore ‘going along’ with surgery as this is viewed as being routine (Acum, 2018).’The literature therefore supports the narratives of clinicians in this study, in that young people may present to surgery, feeling that this is the ‘next step’ in their care, creating feelings of discomfort and internal conflict for professionals. The desire of clinicians to promote SDM and for young people to express a degree of certainty around their motivations for surgery, however, sits in contrast to the experiences of young people, who may feel that taking this position, is ‘burdensome’ and therefore they wish to delegate this responsibility.

Clinicians acknowledge the shift in paternalism, often adopting a view that this should be avoided in order to promote SDM. It is however noted that patients may value clinical paternalism or authoritarianism in some contexts, as this may reduce uncertainty, unpredictability and may relieve the patient of taking responsibility for understanding and responding to a frightening situation (Cole, 2013). This therefore speaks to the narratives shared by clinicians, around balancing patient autonomy and beneficence.

In responding to this ethical dilemma and experience of discomfort, many factors were found to mitigate these experiences. Feeling held and contained within the MDT, and the importance of being able to have ‘ad-hoc’ discussions and opportunities for liaison was felt to be a facilitator to the decision-making process. Similarly, clinicians felt the input from clinical psychologists was particularly beneficial in trying to understand a young person's motivations for surgery, or when questions arose around a young person's capacity to make an informed decision.

Previous research suggests that clinicians do not feel they have the right environment, knowledge, or education to address psychosocial factors with their patients (Clarke and Cooper, 2001; Rumsey et al., 2004). Consistent with this, clinicians highlighted how the SDM environment, where the patient is joined by multidisciplinary professionals in one room, can be intimidating and may pose a barrier to SDM. This was also reported by young people in sharing their experiences of decision-making (Acum, 2018). Clinicians acknowledge that patients may experience dissatisfaction with their appearance, underpinning motivations for surgery, however this can sometimes be difficult for clinicians to elicit due to the nature of the environment in which the discussions around OS are had. Clinicians spoke of reducing the number of professionals in the room and trying to mitigate power dynamics by making modifications to the environment that promote SDM. Due to this, it is often the case that patients share information in their consultations with individual MDT members, highlighting the importance of professionals having opportunities to liaise with the team.

Socioeconomic factors have been reported as a barrier to accessing healthcare, in which financial implications associated with travelling to appointments have been found to impact attendance (Stock et al., 2018). Consistent with this, clinician narratives highlight several barriers to accessing care, including the need for patients to travel to different sites and the associated financial, education and employment considerations that this involves.

Although be-spoke clinics offer opportunities to access care closer to home, in accordance with the CSAG recommendations, clinicians voiced inequity between services, describing this as a ‘mixed economy’, which may contribute to health inequalities.

Clinicians were mindful that patients may experience co-morbid mental health difficulties and were conscious of not proceeding with OS when this was unlikely to meet the patients’ expectations. Social media platforms were noted as a significant influence on patients’ perception of themselves and their appearance and during the SDM discussions clinicians were aware of not ‘biasing’ the young persons decision or creating feelings of dissatisfaction in their use of language.

Clinician awareness and consideration of cultural factors differed across professional groups, with some professionals sharing that these considerations had not arose in their work and could be considered more by the team. In contrast, where cultural factors were acknowledged, this centred around paternalistic approaches and accessing an advocate in managing decisional conflict.

Critical evaluation

The empirical study benefits from having representation from three different professional groups working in cleft services across a variety of geographic regions. Professionals were therefore able to speak to the similarities and differences in the set up of their teams and acknowledged to a lesser or greater degree, health inequalities impacting on access to care. Professionals were also consulted during the set up and design of the project, including seeking feedback on the interview topic guide. Clinician feedback on taking part in the study was largely positive, bringing awareness to different areas of their work that they may wish to consider further with their teams.

The chief investigator (HC) was aware of their role as a Trainee Clinical Psychologist throughout the study and was mindful that this position may have influenced participant disclosure around psychological factors or narratives shared around the role of psychology within teams. In being aware of this, a reflective diary was kept, in enhancing reflexivity and considering positionality and influence during the course of the project.

It is acknowledged that only five out of a possible 12 cleft teams partook in the study, largely due to the scope of the thesis project and time constraints, however due to the nature of how some of the included cleft centres are set up and the role of different professionals within these teams, clinicians had involvement or worked in collaboration with other centres. Owing to the reported differences across teams, the findings should be considered in the context of how teams operate and their unique qualities.

Clinical implications

The need for commitment associated with making a decision to undergo OS is highly recognised in previous literature and by clinicians who participated in the study (Acum, 2018). Having an awareness of the factors that may facilitate or pose a barrier to the decision-making process is paramount, which includes considering the impact of health inequalities, communication style, the provision of information and being aware of psychological, social, and cultural factors that may influence decision-making.

At the transition period in particular, it is important to consider how the delegation of responsibility in decision-making is experienced by young people, and to have an awareness of external influences that may influence decision-making; such as the impact of accessing social media, online communities, and experiencing pressure from support systems.

The environment by which young people and clinicians navigate the decision-making process also warrants attention, of which it is important to consider power dynamics and psychological safety in eliciting motivations for surgery and reaching a ‘collaborative’ decision. With this in mind, having regular opportunities to liaise, discuss and seek support from colleagues in the MDT may enable SDM and reduce feelings of discomfort, particularly for those that are positioned as the final decision-maker. It may also be beneficial, to foster opportunities for liaison earlier on in the treatment pathway, in paying particular consideration to the transition period and experiences of young people at this life stage.

Further research

Although the present study brings awareness to health inequalities that may preclude patients from accessing services, as this was not the focus of the empirical project, this arguably provides a limited account of these experiences. As noted by professionals, this is a topic that understandably warrants further research and resources to be allocated. Developing a greater understanding of the factors that may preclude patients accessing care, and increasing awareness of health inequalities, will allow services to consider how these may be addressed. Further research may also wish to explore how teams consider and incorporate SDM models at different stages of the treatment pathway.

Conclusions

The current study provides insight into the decision-making experiences of clinicians and highlights how awareness of psychological, social, and cultural factors may differ amongst professionals and may influence the decision-making process. The findings also bring awareness to aspects of team working and health inequalities that may impact on cleft care, warranting further discussion and consideration of the factors that may preclude patients attending clinic.

Chapter Five: Extended methodology and reflexivity

The following chapter will provide supplementary information regarding the empirical projects design and methodology, researchers epistemological stance and will provide a discussion around personal reflexivity and ethical considerations.

The empirical study employed a qualitative design in exploring the experiences of clinicians involved in surgery decision-making. Merriam (2009) in describing the use of interpretive and constructivist approaches, defines these as being designed to study the "multiple realities, or interpretations, of a single event" (p9). In accordance with interpretive-constructivist approaches, adopting this methodology and stance allowed the researcher to gain a rich, in-depth understanding of the individual experiences of different MDT professionals in relation to their specific roles within the cleft MDT, but also provided insight into the personal perspectives of individual participants in supporting young people in their decision-making for orthognathic surgery.

Previous craniofacial research has utilised qualitative methods in exploring the experiences of young people and their families (Acum, 2018; Safarikova, 2021) of which it is highlighted that qualitative methods may be more appropriate when considering the challenges associated with recruiting larger samples (Stock et al., 2018). This is particularly applicable, when considering the structure and intimate relationship of cleft MDTs, of which there may be a limited number of professionals available to interview from each discipline.

Eligibility Criteria

Participants were recruited if they were a Speech and Language Therapist (SLT), Orthodontist or Surgeon currently working in a Cleft MDT at one of the approved NHS sites.

Clinicians were required to have experience of being involved in the decision-making process for orthognathic surgery and 3 clinicians from each professional background were recruited. Clinicians needed to have access to Microsoft Teams and consent to audio and video record for the purposes of transcription was also required. Clinicians were not able to take part if they worked outside the NHS or if they worked for a private organisation.

Selection of participants

Participants were recruited via purposive sampling with 9 participants being recruited across 4 sites. The nature of the project being a 'multi-site' project, meant that professionals taking part, represented different geographic areas and cleft teams. A Principal Investigator (PI) was appointed at each individual site, who took responsibility for disseminating a participant information sheet (PIS) and email template to their respective clinical teams. The PI's role was essential to the recruitment of clinicians, in that the PI's were in most cases, embedded in their clinical teams and able to have direct discussions with their colleagues about taking part in the study. Upon clinicians expressing an interest in taking part, the Chief Investigator made contact with the participant directly by email, ensuring that a copy of the PIS had been received, and that any questions were sufficiently answered. A date and time to meet for an interview was then arranged via Microsoft Teams. A consent form was completed and returned by email prior to interviews commencing (Appendix 16) and the researcher provided further opportunities to ask questions at the beginning of the interviews. Additional verbal consent was gained in relation to recording and transcribing interviews.

Data Collection

Interviews were conducted remotely via Microsoft Teams at a time convenient for clinicians. 90 minutes were allocated for each interview, which included time to answer questions, obtain consent, and offer a debrief at the end of each interview. All clinicians took up the opportunity to reflect on taking part in the study, of which they expressed curiosity regarding dissemination and implications resulting from the project and spoke about the benefits to taking part. A semi-structured topic guide was implemented, which was developed and reviewed by a Clinical Psychologist, Surgeon, Speech and Language Therapist and Orthodontist working in cleft settings prior to the study commencing (See Appendix 12).

Ethical Approval

Ethical approval was obtained from the University of East Anglia and Health Research Authority (HRA). Subsequent local approvals were obtained from the research and development teams (R&D) at 5 participating NHS sites. Research was conducted in line with the British Psychological Society (BPS) code of ethics and conduct (BPS, 2018). During the initial stages of designing the project, the Chief Investigator (HC) and primary supervisor had the opportunity to attend a Clinical Excellence Network (CEN) meeting which was attended by Clinical Psychologists working across cleft services in the UK. The Chief Investigator and Primary Supervisor also presented at the East of England MDT cleft meeting. The project received a favourable opinion from MDT members across both groups, in which CEN psychologists gave their support for the project and Speech and Language Therapists advised of including their professional group in the study.

Informed consent

The PI for each site, first made contact with the clinicians working in their team through disseminating an email template and PIS. Following this, clinicians directly expressed an interest in taking part in the study and were followed up accordingly. Written consent was obtained prior to the interviews commencing and professionals consented to being audio and video recorded for the purposes of transcription.

Confidentiality

Anonymity was maintained throughout the transcription process through assigning participant ID''s to each participant. Personally identifiable information was removed from interview transcripts and data was stored securely on UEA OneDrive, adhering to the GDPR regulations (Information Commissioner's Office, 2018)

Distress

It was possible that the nature of discussing ethical dilemmas and aspects of the decision-making process for surgery, may have been challenging for clinicians. Clinicians were offered a debrief at the end of their interview, in reflecting on their experiences of taking part in the study. Although the need to access further support did not arise during the study, options for further support would have included seeking support from within their team from the cleft team psychologists.

Risks/Benefits/Burdens

It is acknowledged that taking part in an interview for the study, required clinicians to protect 90 minutes of their time. Understandably, with the pressures facing many clinical teams within the NHS, this may have contributed to workload. In obtaining capability and capacity approval from the individual R&D departments, it was deemed that the clinical teams had capability and capacity to accommodate the study, and the chief investigator was happy to remain flexible and accommodate interviews on a day and time suitable for clinicians. Feedback from the interview process was largely positive, with clinicians sharing that this provided opportunities to reflect, and to consider different themes in relation to their work. Clinicians were also offered a £10 Amazon e-voucher as a token of appreciation for taking part.

Thematic Analysis (TA), positionality and reflexivity

Data was analysed and interpreted using Thematic Analysis (TA), in which the guidelines for conducting Thematic Analysis outlined by Braun and Clarke (2006) were followed. Thematic analysis was chosen due to the flexibility in its approach, which means it does not stem from a specific epistemological or theoretical position. This approach, as noted by Braun and Clarke, is therefore compatible with a constructivist position, and is also considered to be less governed by rules during the analysis phase. This allows for more in-depth meaning of the data can be obtained, in which theory is therefore derived from the data itself in line with the exploratory aims of the research.

Reflexivity is commonly practiced in the context of undertaking qualitative research and is well acknowledged within academia. The process of reflexivity involves the researcher reflecting on their own positionality and influence in relation to the research being conducted, in which the purpose of reflexivity is intended to 'evaluate' or 'measure' the quality and rigour of qualitative studies. Braun and Clarke (2019) in publishing a reflective commentary on the topic of 'reflexive thematic analysis', identify that 'themes do not passively emerge from either data or coding' instead, Braun and Clarke (2019) argue that 'themes are creative and interpretive stories about the data, produced at the intersection of the researchers theoretical assumptions'. It is therefore imperative for researchers to acknowledge their positionality and theoretical position in respect to the interviewing process, but also throughout the process of coding and conducting TA. In the following section, the researcher addresses personal reflexivity and use of a reflective diary in paying due attention to positionality and influence.

Personal reflexivity

Lived experience and EDI involvement.

As a trainee who identifies as having lived experience of accessing services, I am particularly passionate about equality and diversity, and improving patient experience (EDI). As such, I have also been involved in supporting EDI projects at the university during my training journey. Exploring how clinicians encompass psychosocial and cultural factors within surgery decision-making, is therefore a topic area that closely aligns with my own values and interests. With this in mind, I am mindful that my specific passion for EDI and exploring psychosocial and cultural factors, may potentially influence what aspects of conversations I become attuned to, during the interview process and in coding. Conversely, being involved in EDI projects, and increasing my awareness of issues of equality and diversity, may have positively influenced the follow up questions that were asked, in gathering further information related to particular topics, and may also have also increased my awareness of these themes within the process of conducting my analysis.

In further reflecting on my own experiences and position in relation to the research, I am also an individual who has received orthodontic treatment during childhood and who has also undergone surgery at various stages of life, spanning school age to adulthood. During this time, I also attended various health appointments during my primary school years, which involved taking time off school and liaising with different health professionals. In considering these experiences, I have reflected on how there has been a significant shift in decision-making, particularly around the involvement of young people in SDM and ensuring that young peoples voices are heard. It is also possible that these experiences, may influence my attunement to certain narratives around shared-decision-making, particularly the involvement of young people within the shared decision-making process for surgery and how this is experienced with regards to psychosocial and cultural factors. In considering this position, I used a reflective journal throughout the project, which allowed me to consider my own role in relation to the research. Codes and themes were also discussed in supervision, in which sections of a variety of anonymised transcripts were also reviewed by the research team (KM and EY) in improving quality and rigour.

Role as a Trainee Psychologist

During discussions, I was mindful that participants would be aware of my role as a Trainee Psychologist. As the participant sample consisted of clinicians working alongside Clinical Psychologists in their role, it is highly likely, that this influenced the interviewing process. Indeed, this may have influenced the narratives shared around Psychology as a discipline, or discussions centred around interactions with psychologists. Similarly, when discussing psychosocial factors, this may have influenced the way in which psychological factors were discussed. This was considered during the process of building the interview topic guide, in which questions were purposefully framed openly around the factors considered during decision-making, prompting around psychological, social, and cultural factors depending on the conversation and what was elicited. The interview topic guide was also reviewed by a Surgeon, Orthodontist, Speech and Language Therapist and Clinical Psychologist working in cleft services, in ensuring the questions felt representative.

Impact on clinical practice

The experience of interviewing clinicians has been an invaluable process. During the course of the thesis project, I had joined a paediatric psychology service, of which the themes surrounding health inequalities and psychosocial and cultural factors have also become apparent in my own clinical work. It is therefore important to highlight, that the themes raised in this study, are not only applicable to cleft surgery decision-making, but are also applicable to professionals working in paediatric health settings more widely and EDI projects conducted within services.

Chapter Six: Extended Results, Discussion and Critical Evaluation

The thesis portfolio aims to provide an overview of the reported barriers and facilitators to shared decision-making in paediatric surgery, with a focus on exploring the decision-making experiences of clinicians in the context of orthognathic surgery in cleft services.

Across both papers, the importance of the patient-physician relationship, building trust and rapport and the provision of timely and reliable information, is acknowledged in promoting shared decision-making in paediatric surgery (Atsaidis et al., 2022; Papiez et al., 2021; Lecouturier et al., 2015). Conversely, receiving too much, or too little information, and paternalistic approaches have been reported to pose a barrier to engaging in SDM (Samanta et al., 2022; Atsaidis et al., 2022; Sjoberg et al., 2015; Heath at al., 2016)

The importance of considering psychosocial factors and cultural and religious values within SDM is highlighted in the literature, of which a cleft may be viewed by some communities as being a ‘gift from god’ or resulting from supernatural or mythical forces (Hasanuddin et al., 2023). Clients may also report experiences of stigma in their communities, which has been associated with scarring and concerns around finding a marriage partner (Stock et al., 2018). Having an awareness of cross-cultural beliefs is therefore paramount to promoting positive health outcomes and collaboration, however the findings of the empirical study suggest that there are still health inequalities that need to be addressed, that preclude patients turning up to clinic. These include, but are not limited to, financial considerations, expectations around accessing a dentist and having “good oral health”, child-care considerations and educational/employment implications. In addition to this, when presenting to clinic, clinicians report experiences of uncertainty with regards to accessing interpreters or felt that cultural considerations did not arise in their work or could be considered further.

Additional results will be provided below, which could not be captured in the empirical paper due to the word limit set by the chosen journal. The current chapter will also provide a summary and critical evaluation of both papers, discussing the findings and clinical implications in relation to the evidence base and wider theoretical context.

Summary of key findings

Taken together, the findings of the systematic review and empirical paper provide an overview of different factors that clinicians may experience or report whilst navigating the shared decision-making process for paediatric surgery and orthognathic surgery specifically. Key findings from both papers will be discussed, making reference to the evidence base and theoretical implications.

Systematic Review

A systematic review was conducted with the aims of exploring the perceived barriers and facilitators to shared decision-making, as reported by young people, their families and/or caregivers and clinicians involved in the decision-making process for paediatric surgery.

Seventeen papers were included in the final report, of which four analytic themes and 11 major descriptive themes were identified. Interpersonal qualities, communication, and considering sources of support, were all acknowledged as factors impacting on decision-making across both projects. In particular, having the opportunity to revisit discussions with the clinical team and being given sufficient time to make decisions for paediatric surgery, were identified as factors that enable or facilitate shared decision-making (Atsaidis et al., 2022; Samanta et al., 2022) Being provided with reliable information that is tailored to the young person was also reported to be a facilitator to shared decision-making, of which families reported to find it helpful when statistics or percentages were shared (Carlisle et al., 2022; Loeff and Shakhsheer., 2021)

Interpersonal qualities of the clinician and the relationship with the young persons paediatrician in particular, were found to be facilitators to the decision-making process. Families reported to find it beneficial to include their child's paediatrician in SDM discussions (Boss et al., 2017; Waite et al., 2022), however it is acknowledged that this is predominantly reported in American based studies, whereby the paediatrician may hold different roles and responsibilities. Empathy, kindness, and genuineness of clinicians were reported to enable SDM, of which using lay language, using a slow conversational pace, and increasing involvement of families in SDM, were reported as facilitators (Carlisle et al., 2022). Conversely, families not feeling listened to or involved in SDM, receiving too little or too much information, use of 'medical jargon' and perceiving apathy or judgement from clinicians were reported as barriers to engaging in SDM (Samanta et al., 2022; Smith et al., 2013; Links et al., 2020; Waite et al., 2022).

Empirical Paper

A qualitative study was employed in exploring clinicians understanding of psychological, social, and cultural factors and how these factors may be considered during the decision-making process for orthognathic surgery. The study involved conducting 9 interviews with 3 speech and language therapists, 3 Orthognathic Surgeons and 3 Orthodontists involved in the decision-making process at the transition period, of which the results of a thematic analysis revealed 3 major themes and 7 subthemes. Clinicians reflected on the complex nature of balancing patient autonomy and acting in the best interest of young people, which created a sense of internal conflict and discomfort for professionals who were positioned as the ‘final decision maker’ for surgery. Having opportunities for liaison with the multidisciplinary team and having the valued contribution of different team members to the decision-making process, were reported to be helpful in managing feelings of discomfort and were found to facilitate decision-making.

The environment by which professionals and young people navigate the decision-making process, however, was acknowledged as a potential barrier to engaging in decision-making. When consulting with young people and the multidisciplinary team about orthognathic surgery, power dynamics were acknowledged as potentially hindering shared decision-making, with consideration being given to the number of professionals in one room and how the chairs and dental chairs are positioned. Health inequalities and perceived equity across services were also raised as barriers to accessing cleft care, precluding decision-making for orthognathic surgery.

Extended results

Awareness of psychological and cultural factors: In asking clinicians about any cultural factors, they consider during the decision-making process for orthognathic surgery, awareness and consideration of cultural factors differed across professionals. For some, specific examples were shared around working with patients from different cultural and religious backgrounds as referenced in the empirical paper. In contrast, other professionals felt that cultural factors had not arose in their work, or descriptions of working with cultural factors was limited to referencing collaboration with interpreters or considering timing of surgery around religions holidays.

> Maybe we don't. I don't know. specifically cultural... I'm not sure we do address that particularly well (Participant 1).

> in my limited experience, I don't think I've come across any patients with whom I thought…or that it's come up that there's any cultural factors That might have impacted on the care that I would give… other than and possibly having to involve an interpreter.' (Participant 8).

> I can't think of any particular examples where. uh, where a patient's ethnicity may have changed our relationship with them or change the way that we manage the MDT because our aim is, is to provide the same service for every patient. So, you know, I'm sure there are considerations around, you know, I don't know. Um, but you know, if you want your jaw surgery on Christmas Eve or if you want it on Eid or you know, whatever, I think I think we would we we're completely flexible about those sorts of things (Participant 6).

> I am not thinking of a moment where any of those have, like any cultural…social or psychological... factors, have sort of come up. I'm sure it would... come up if. Psychology appointments but yeah, I don't know any that come up. I don't think I can't think of an example anyway. (Participant 9)

In speaking with professionals, it is acknowledged that often limited to no training is provided around understanding psychological or cultural factors, supporting the findings of Stiernman et al. (2019) who reported that health professionals often derive their knowledge from conferences or seminars as opposed to receiving formal training. These findings may therefore have implications for clinicians in receiving more training around psychosocial and cultural factors.

Barriers and facilitators to shared decision-making in OS.

In considering factors that are perceived to enable or pose a barrier to the decision-making process, clinicians made reference to use of language, humour, acceptability of returning to the treatment pathway, emotional maturity and comprehension level. Clinicians were also mindful of the need to give time to consider decision-making, and clinicians acknowledged factors such as educational background and presence of learning difficulties that may require adaptions to SDM.

> I always try and make a little joke, you know, like, nobody else looks at this type of level, like, I'm just a big nerd and I'm doing all my measurements and I almost say something like self deprecating to not make them feel bad that I'm doing things like pretty critical measurements.' (Participant 5).

> Parking people with their thoughts for a while is probably my most commonly used tactic… if I feel either, the patient needs a bit more time to think... to digest things…or if I need...if I'm not happy and I think that the possibly someone's trying to get something that is maybe not going to work for them' (Participant 7).

> So that would make her MDT appointment slightly different from, you know, another 16-year-old that. Have a very similar. Jaw set up very similar, you know, potential improvement but has family support, comes with mum and Dad has just finished GCSEs is on a college…You know all those sorts of subtle differences and

that that young person. In the second sort of group would be able to understand written information. Would be able to go away and reflect and have the family support to discuss things at home, whereas the young lady probably won't. So… it's about it's about how we tailor that.’ (Participant 3).

It's never off the table. And even I said I say to patients like if you say you don't want surgery, I'm not going to be mad. Like, I'll have this conversation with you later. It's never off the table, you know. So that they know that you know it's not a black and white decision. So, if they do decide when they're 25 that they want to have this done, then come back and have a chat. So, I think the door is kind of always open and that's a really good MDT’ (Participant 5)

And I think that actually that's something that our service is really good at and we always try and let patients know that actually this doesn't have to be something that you have done now, this can be something that is, you know, years in the future or even decades in the future if you ever decide that you wanted to pursue it once it and I do think that we're really good at saying that and providing that as a service that if anybody wants to come back into the service, we're always very happy to do that.’ (Participant 8)

I think the young person's...emotional wellbeing, their maturity, their comprehension. Because I think we do have some young people with some learning needs, and I think understanding risks versus benefits can be much harder for them.’ (Participant 3).

These findings support those reported in the systematic review, of which gauging comprehension level and having limited language or comprehension skills were reported to influence the shared decision-making process (Atsaidis et al., 2022). Similarly the importance of giving time to make a decision and offering repeated visits and opportunity for discussion, are acknowledged in both the systematic review and empirical study (Almoajil et al., 2022; Timmermans et al., 2018; Samanta et al., 2022; Papiez et al., 2021).Furthermore, the results also support those of Kleinman (1979) in which the importance of considering a patients comprehension level, educational background and language is highlighted.

Thesis strengths and limitations

Systematic review: A strength of the systematic review, is that the report encompasses a variety of different study types, including a scoping review, mixed methods study, observational study, and case report in addition to qualitative studies. The review also includes research undertaken internationally, such as in the USA, Canada, and Sweden.

Due to the limited published research available around the barriers and facilitators to engaging in shared decision-making in paediatric surgery, including a variety of study types and keeping the inclusion criteria broad allowed for a more comprehensive overview of the literature. The systematic review also benefits from having a secondary screener during the screening of titles and abstract stage, at the full text screening phase and in quality appraising studies, enhancing the quality and rigour of the review. Studies were also critically appraised respectively using different quality appraisal tools, owing to the differences in methodology.

Although the review included a variety of study types, and were appraised accordingly with specific appraisal tools, greater difficulty was experienced in appraising observational and case report studies. It is reported that critical appraisal tools vary in their intent, components, construction, and psychometric properties, therefore in appraising research, consideration should be given to the properties and appraisal tool chosen (Katrak et al., 2004). Although the appraisal tools selected to appraise observational research and case reports were assessed to be the most appropriate, these tools did not adequately represent the quality of the studies, receiving a lower rating due to questions being ‘non-applicable.’ These papers were discussed in research supervision, of which the decision was made to include these papers, due to their valued contribution to the review. A further limitation that was considered, is that the review did not include grey literature, of which further reviews may wish to include unpublished or grey literature in addressing publication bias.

Empirical Paper

Previous research has reported on the experiences of young people and their parents in making decisions for orthognathic surgery, with suggestions for further research centring around exploring the experiences of clinicians involved in the decision-making process. This study provides an in-depth exploration of the decision-making process at the transition period, with a focus on understanding how Clinicians understand and consider psychological, social, and cultural factors when supporting young people in their decision-making for OS.

The study benefits from having the participation of clinicians in designing the interview topic guide and having the opportunity to role play the interview topic guide with a professional working in the field beforehand. This allowed the researcher to consider the impact asking different questions would have on participating clinicians, specifically when considering how to introduce questions around ethical dilemmas. Role playing the interview topic guide, also provided an opportunity to develop prompting questions further.

As the systematic review was conducted prior to the empirical study commencing, the researcher had an awareness of different barriers and facilitators being reported in relation to paediatric surgery decision-making. The researcher continuously reflected on the potential impact of having this knowledge on the interpretation of the findings and generation of themes from the data. Themes were discussed in supervision, of which the researcher reflected on any potential influence with the research team.

Theoretical implications

In contextualising the findings of the systematic review and empirical paper using the model of shared decision-making proposed by Park and Cho (2018), the theme 'navigating the decision-making process' and clinicians speaking to the difficulty of balancing patient autonomy and beneficence aligns strongly with this. In hearing clinician narratives, particularly those of clinicians who are positioned as 'final decision maker' for orthognathic surgery, the desire for certainty in the decision-making process became apparent. Clinicians acknowledge the shift in paternalism, of which they are aware of the importance of involving the young person in shared decision-making, however this can sometimes be difficult for clinicians and create feelings of discomfort when motivations for surgery are not clear or when young people present to surgery having been 'told' orthognathic surgery is required or the natural next step in the treatment pathway. Clinicians therefore may find themselves in a position whereby complete certainty cannot be obtained, therefore reaching a 'joint decision' or 'compromise' that both parties can accept and come to an agreement on. This decision will be reached upon the outcomes, treatment benefits and risks having been discussed with the child or young person and their family, as outlined in Park and Cho's model (2017). In exploring motivations around surgery, this will also involve identifying a ‘common goal,’ of which clinicians spoke about the valued opinion of MDT members, such as that of psychology in exploring motivations for surgery, or speech and language therapists in advising of any changes to speech. Clinicians spoke of the importance of understanding the young persons motivations for surgery and managing expectations in order to be able to proceed with decision-making for surgery, aligning with the attributes listed in Park and Cho’s model.

In contextualising the findings of the systematic review using Park and Cho’s model, Park and Cho highlight how shared decision-making requires the active participation of all parties (child, parents, and Clinicians). In the systematic review, parents not feeling involved in decision-making, not feeling listened to, or clinicians perceiving families or young people as not wanting to be involved in SDM, were reported to be barriers to SDM. Not perceiving families or young people as wishing to be involved in SDM, may also speak to the ‘antecedents’ listed in Park and Cho’s model, of which ‘willingness to participate in decision-making’ is listed as an antecedent to paediatric decision-making. Several barriers and facilitators identified around use of language, communication and interpersonal skills, provision of information, and identifying support around the family and young person can be viewed as impacting on ‘collaborative partnership’, which also focusses on understanding the families’ values. As reported in the systematic review, considering the families values, beliefs and preferences were also noted as being a facilitator to SDM (Atsaidis et al., 2022) aligning with the model proposed by Park and Cho (2018).

Jordan et al. (2020) in reporting on the viewpoints of a paediatric patient, parent, and paediatrician about the use of SDM in paediatrics, highlight the importance of the patient-physician relationship, building trust and valuing patient experience and knowledge in facilitating SDM. The young person shares their experience of being provided with advice that was counter to what their experience told them was best. They also shared that having increased knowledge about their care and treatment, enabled them to feel more comfortable asking questions and increased their engagement in treatment.

The results from the systematic review and empirical paper, also make reference to these themes. In the systematic review, receiving contrasting information, not feeling listened to, and apathy to engage in open dialogue were reported to pose a barrier to SDM. Similarly, providing consistent information and building a trusting relationship with patients were found to enable SDM.

In providing a parent's perspective on engaging in SDM, the parent shares that the unfamiliarity of the symptoms and medication effects could feel intimidating and made it difficult to get involved in decision-making. In support of this account, the findings of the systematic review, suggest that it is beneficial for clinicians to provide preparatory information and support parents in seeking out health information, signposting to peer support and sources of reliable information. Assuming parental knowledge and comprehension level, was also reported as a barrier to engaging in SDM, in support of this account. These findings are also supported by the empirical paper, in that clinicians spoke of signposting patients to 'reliable' information (for example, from the British Orthodontic Society) or to peer information.

In agreement with the young person's account of engaging in SDM, a Paediatrician reports that a facilitator to this process is having a longstanding relationship with the patient, obtaining a history to build upon in understanding their goals. They further share that this has allowed them to develop a 'partnership' with families, in which they are aware of the intention to make decisions together. Conversely, if a patient is new and there is limited time to discuss treatment options and facilitate SDM, this may pose a barrier to engaging in the process. Across clinician narratives in the empirical study, themes also emerged around the importance of the patient-physician relationship. Orthodontists were remarked on as having a longstanding relationship with patients across the cleft treatment pathway, of which they often have an insight into the patient's life, their thoughts and personality. In contrast, being unknown to the patient, was reported to pose a barrier to shared decision-making for orthognathic surgery in the sense that the clinician is attempting to explore motivations for surgery in a setting whereby therapeutic rapport and psychological safety are yet to have developed and are in some cases, adversely affected by the shared decision-making environment. This also provides evidence in support of the young person's account, highlighting the importance of the patient-physician relationship and rapport building.

Leadership in the NHS

As the NHS continuously evolves, there has been a shift from 'command and control' structures towards adopting collective, inclusive and compassionate leadership styles (Bailey and West., 2022) In implementing this approach, there is a greater emphasis on distributing leadership to where expertise, motivation and capability lies and avoiding imposing decisions from the top down. This is also re-iterated in the NHS Long Term Plan (NHS England, 2019), whereby one of the aims is to 'strengthen and support good, compassionate, and diverse leadership at all levels. In accordance with this, when interviewing clinicians about their decision-making experiences around orthognathic surgery, clinicians spoke about everybody being a 'clinical lead' and having a role to play within the shared decision-making discussion with patients. One clinician highlighted how it was important that the team was not seen as being ‘surgeon led,’ and meetings were often facilitated by professionals who had the most input or experience with the patient, speaking to their speciality and input.

Adopting a compassionate leadership style, is also understood to include listening to staff and arriving at a shared understanding of the challenges they face. In the empirical study, this was felt to vary depending on the set up of teams, and whether these were centralised or bespoke. Although clinicians reported to have strong working relationships with their colleagues, including building links with community colleagues, there was a desire for more opportunities for liaison. Ad-hoc conversations were remarked on as being beneficial for clinicians, however for some teams, there was variance in equity and health inequalities across services, with some teams feeling that more opportunities for liaison would be beneficial.

Participant feedback on the interview process

Positive feedback was received regarding taking part in the interview process, with clinicians sharing that this provided opportunities to take a step back from the decision-making process and ‘look in.’ In particular, clinicians commented that the interview processes provided an opportunity to consider aspects of equality, diversity and inclusion and to bring ideas back for discussion with the clinical team and clinicians also felt that the interview process allowed for greater consideration of the transition period specifically, bringing awareness to factors that may be beneficial to consider in relation to this. Positive feedback was also shared from the Clinical Excellence Network meeting, in which the provisional results from the study, were shared in February 2024.

Health Inequalities

The prevalence of health inequalities impacting on access to cleft care are highlighted across the literature, with socioeconomic status and identifying as being from a marginalised background being reported as factors impacted on access to care (Smillie et al., 2015; Stock et al., 2018; Abbott et al., 2011; Zaluzec et al., 2019).

In a study exploring clinical directors views of centralisation and commissioning of cleft services in the UK (Searle et al., 2015) varying accounts regarding equity in services and access to care were reported. In one account, the participant gives the opinion that the team is ‘team centric’ whereby the cleft team rotate around the patient, rather than patient and staff rotating around the surgeon, speaking to the advantages of moving towards greater centralisation of teams.

In contrast, regional differences were highlighted with a participant sharing the need for ‘equity of cleft care’ and that ‘every centre has a different model of funding’ highlighting disparities. Another participant also raised how clinicians may be required to travel to and between services, which could have an adverse effect on staff and lead to ‘burn out’.

In the empirical study, concerns regarding equity in services and barriers to accessing healthcare were also reported by some clinicians. Clinicians spoke of how professionals in attendance at different clinics may vary and ‘obstacles’ that preclude patients turning up to clinic, such as the requirement to see a dentist regularly and have ‘good oral health’ amongst employment, financial or childcare considerations were also noted. A clinician remarked on how they don’t see the same mixture of backgrounds in their clinic, that they would expect from the general population, suggesting there is still a need to address health inequalities impacting on access to care that need to be addressed, prior to reaching discussions around orthognathic surgery. The evidence base suggests that working in a more synchronised manner and increasing information sharing, may be helpful in addressing health inequalities and improving practises (Wagner et al., 2021). supporting the themes raised in this study around fostering team liaison.

Research implications.

In combining the findings from both the systematic review and empirical study, the portfolio highlights two key areas for further research. Shared decision-making is a well-established concept within the field of surgery decision-making, however much of this research has focussed on adult populations. It is recommended that further research focusses on reviewing the literature in this field, which may include evaluating the impact of shared decision-making interventions in paediatric settings in particular. Furthermore, the portfolio brings awareness to health inequalities impacting on access to care, precluding shared decision-making being able to take place. In meeting the six core principles that underpin our NHS values, it is therefore of significant importance to conduct further research to explore and address health inequalities impacting on access to cleft care.

Overall conclusions

Overall, the thesis portfolio highlights a complex interplay of factors that impact on the decision-making process for paediatric surgery. Areas for particular consideration centre on developing a greater understanding of health inequalities impacting on access to cleft care and considering the different ways in which teams can enhance communication and liaison in services where there is a desire to increase this or whereby less opportunities to access these spaces are reported. Services may also wish to focus specifically on factors emerging at the transition period in particular, sharing knowledge around barriers and facilitators impacting on the decision-making process at this specific life stage whereby the set up of services and organisational structures may mean this is more or less of a transition for young people (e.g. from children to adult services or remaining in the same system).

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Appendix 18. Example Theme Development

Appendix 19. Visual representation of searching for themes and subthemes

Appendix 20. Worked example of reviewing and defining themes

Appendix 1: Author Information, The Journal of Pediatric Health Care (JPHC)

Author Information

Introduction

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Inclusive language acknowledges diversity, conveys respect to all people, is sensitive to differences, and promotes equal opportunities. Content should make no assumptions about the beliefs or commitments of any reader; contain nothing which might imply that one individual is superior to another on the grounds of age, gender, race, ethnicity, culture, sexual orientation, disability or health condition; and use inclusive language throughout. Authors should ensure that writing is free from bias, stereotypes, slang, reference to dominant culture and/or cultural assumptions. We advise to seek gender neutrality by using plural nouns ("clinicians, patients/clients") as default/wherever possible to avoid using "he, she," or "he/she." We recommend avoiding the use of descriptors that refer to personal attributes such as age, gender, race, ethnicity, culture, sexual orientation, disability or health condition unless they are relevant and valid. When coding terminology is used, we recommend to avoid offensive or exclusionary terms such as "master", "slave", "blacklist" and "whitelist". We suggest using alternatives that are more appropriate and (self-) explanatory such as "primary", "secondary", "blocklist" and "allowlist". These guidelines are meant as a point of reference to help identify appropriate language but are by no means exhaustive or definitive.

Reporting sex- and gender-based analyses

Reporting guidance

For research involving or pertaining to humans, animals or eukaryotic cells, investigators should integrate sex and gender-based analyses (SGBA) into their research design according to funder/sponsor requirements and best practices within a field. Authors should address the sex and/or gender dimensions of their research in their article. In cases where they cannot, they should discuss this as a limitation to their research's generalizability. Importantly, authors should explicitly state what definitions of sex and/or gender they are applying to enhance the precision, rigor and reproducibility of their research and to avoid ambiguity or conflation of terms and the constructs to which they refer (see Definitions section below). Authors can refer to the Sex and Gender Equity in Research (SAGER) guidelines and the SAGER guidelines checklist. These offer systematic approaches to the use and editorial review of sex and gender information in study design, data analysis, outcome reporting and research interpretation - however, please note there is no single, universally agreed-upon set of guidelines for defining sex and gender.

Definitions

Sex generally refers to a set of biological attributes that are associated with physical and physiological features (e.g., chromosomal genotype, hormonal levels, internal and external anatomy). A binary sex categorization (male/female) is usually designated at birth (""sex assigned at birth""), most often based solely on the visible external anatomy of a newborn. Gender generally refers to socially constructed roles, behaviors, and identities of women, men and gender-diverse people that occur in a historical and cultural context and may vary across societies and over time. Gender influences how people view themselves and each other, how they behave and interact and how power is distributed in society. Sex and gender are often incorrectly portrayed as binary (female/male or woman/man) and unchanging whereas these constructs actually exist along a spectrum and include additional sex categorizations and gender identities such as people who are intersex/have differences of sex development (DSD) or identify as non-binary. Moreover, the terms ""sex"" and ""gender"" can be ambiguous—thus it is important for authors to define the manner in which they are used. In addition to this definition guidance and the SAGER guidelines, the resources on this page offer further insight around sex and gender in research studies.

AUTHORSHIP

The *JPHC* follows the ICMJE guidelines for definition of authorship: "Authorship credit should be based on 1) substantial contributions to conception and design, acquisition of data, or analysis and interpretation of data; 2) drafting the article or revising it critically for important intellectual content; 3) final approval of the version to be published; and 4) agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved" (retrieved from http://www.icmje.org/recommendations/browse/roles-and-responsibilities/defining-the-role-of-authors-and-contributors.html).

Changes to authorship

This policy concerns the addition, deletion, or rearrangement of author names in the authorship of accepted manuscripts:

Before the accepted manuscript is published as an article in press: Requests to add or remove an author, rearrange the author names, or change author credentials must be sent to the Journal Manager from the corresponding author of the accepted manuscript and must include: (a) the reason the name should be added or removed, or the author names rearranged and (b) written confirmation (e-mail, fax, letter) from all authors that they agree with the addition, removal or rearrangement. In the case of addition or removal of authors, this includes confirmation from the author being added or removed. Requests that are not sent by the corresponding author will be forwarded by the Journal Manager to the corresponding author, who must follow the procedure as described above. Note that: (1) Journal Managers will inform the Journal Editors of any such requests and

(2) publication of the accepted manuscript in production is suspended until authorship has been agreed.

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SUBMISSION OF MANUSCRIPTS

All manuscripts must be submitted through Editorial Manager(https://www.editorialmanager.com/YMPH/default.aspx). Authors are requested to submit the text, tables, and artwork in electronic form (not as a PDF). In an accompanying letter authors should state that the manuscript, or parts of it, have not and will not be submitted elsewhere for publication.

Submission of items includes 1) a cover letter, 2) title page with author information, disclosure statement, and key words, (3) the manuscript and references, and (4) table(s)/figure(s) with legends. Note figures and tables are to be submitted as separate files (see below). Revised manuscripts should also be accompanied by a unique file (separate from the cover letter) with anonymous responses to reviewers' comments. Please note that the response to reviewers should not contain any identifying information. The preferred order of files is as follows: cover letter, title page, response to reviews (revised manuscripts only), manuscript file(s), table(s), figure(s).

Papers must be prepared using American Psychological Association (APA) style. You are referred to the Publication Manual of the American Psychological Association, Seventh Edition, copies of which may be ordered from http://books.apa.org/books.cfm?id=4200067 or APA Order Dept., P.O.B. 2710, Hyattsville, MD 20784, USA or APA, 3 Henrietta Street, London, WC3E 8LU, UK.

Manuscripts may be submitted with the goal of offering CE credit (see Continuing Education below).

All correspondence once the manuscript is submitted, including the Editor's decision and request for revision, will be by e-mail.

Preparation

MANUSCRIPT PREPARATION

Manuscripts (text, excluding references and tables) should not exceed 5000 words. Department submissions have varied word counts (see

Department Manuscript Preparation below). If abbreviations cannot be avoided, use the expanded form when first mentioned and abbreviate thereafter. Use generic drug and equipment names (trade names may be listed in parentheses at the point of first mention). If it is necessary to mention a trade name for equipment, the name must be followed immediately by the manufacturer's name and city/state. Pagination should begin with the title page as page 1 and continue through the entire manuscript.

To each page, add Line Numbers, a function of Microsoft Word, prior to submitting. Line numbering is required. Submissions without line numbers will be returned to the corresponding author for correction prior to sending the manuscript to reviewer for comments.

Title Page. Articles require a title page. It should include the title of the manuscript, author names with earned credentials (as per the American Academy of Nursing, http://www.aannet.org), job title, and corresponding author's address, email, and phone number. It should also include any disclosures, acknowledgments, and key words. Note for retired authors: please list your previous position title and institution. If you are also the corresponding author, please use your previous institution's work address unless you would prefer your home address be listed.

Key Words. On the title page, provide 3 to 5 key words, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, 'and', 'of'). Please think carefully about the Keywords for your manuscript. Keywords should be MeSH terms. When your article is published, using the Medical Subject Headings (MeSH) terms as keywords, which are established by the National Library of Medicine, makes it more likely that your article will appear in a search by anyone using medical databases (PubMed, CINAHL, etc) for biomedical or health-related information.

APA format as listed above under manuscript submissions

Research Articles

We welcome all styles of rigorous research investigations addressing the health, disorders, diseases, primary, acute, and specialty care for all ages pediatric, adolescent, and young adults: as well as aspects of practice for pediatric nurse practitioners, and family nurse practitioners advanced practice nurse practitioners focusing on all pediatric populations.

Research Studies include, but are not limited to:

1. **Quantitative studies:** Experimental; Quasi experimental; Descriptive; Correlational

a. Exemplars:

b. Randomized control trials (RCTs) reporting using the CONSORT criteria: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6398298/ OR https://www.elsevier.com/__data/promis_misc/apmr_repguide.pdf

c. Systematic and Meta-analyses reporting using the PRISMA criteria https://pubmed.ncbi.nlm.nih.gov/33782057/

d. https://www.sciencedirect.com/science/journal/26665719/publish/guide-for-authors

e. Integrative reviews: https://www.sciencedirect.com/science/journal/22134220/publish/guide-for-authors

f. Concept analysis: https://pubmed.ncbi.nlm.nih.gov/27450481/

2. **Qualitative Research:** https://www.ncbi.nlm.nih.gov/pmc/articles/PMC8106287/#

a. Phenomenological

b. Grounded theory

c. Ethnographic

d. Narrative

e. Historical

3. **Mixed Methods Research**

a. Reporting guidelines: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC9223457/

4. **Implementation Science:**

a. The Standards for Reporting Implementation Science (StaRI) style

b. https://pubmed.ncbi.nlm.nih.gov/28264797/

-Please refer to the information below for Tables, Figures, and Video data, and Supplementary materials for all research studies.

-Authors for research studies must be able to provide data, if requested by readers who wish to duplicate the study.

-Before authors final submission, please see Elseviers checklist at the end of this author's guide.

Quality Improvement Articles

1. Quality improvement studies in advanced practice pediatric nursing and their families, pediatric health care systems, and organizations are welcomed.

2. Reporting style using the Squire 2.0 guidelines. They may be modified

based on the particular project completed.

3. Squire 2.0

4. Quality Improvement Project articles should include a structured abstract with the following headings:
Background
Local Problem or Aim Statement
Methods
Interventions
Results
Conclusions

Abstract. Abstracts should be limited to 150 words and appear on the first page after the title page. The abstract should be factual, and present the key points in the manuscript, with a summary of clinical implications. Abstracts are published for all full-length articles and some Departments.

Introduction: State the purpose or objective of the study, including the major hypothesis tested, if any.

Method: Describe the study design, the setting, sample, and measures used to collect data.

Results: Describe the major outcomes and statistical significance, if appropriate.

Discussion: State the significance of the results.

Letters to the Editor. Letters to the Editor raising some point of current interest or commenting on an article that appeared in the *JPHC* will be considered for publication. The Editor reserves the right to accept, reject, or excerpt letters without changing the views expressed by the writer. The author will have an opportunity to reply to the comments.

DEPARTMENT MANUSCRIPT PREPARATION

Department features are published in varying frequency. Queries regarding department submissions can be sent to the corresponding or department editor listed. Suggested words counts for manuscript length are listed in the department descriptions. Authors should follow the instructions for full length articles.

Primary Care Case Report. This section features case presentations reflecting either common or unusual clinical situations seen in primary care. The anonymity of patients presented should be maintained and permission obtained from the patient and or family if the patient is not de-identified. IRB approval for the presentation of case reports may be required and is the author's responsibility. Any author interested in sending a query should direct their cases to the Corresponding Editor at joannserota@msn.com. See the suggested template for case report preparation. The suggested word count is 3000.

Reporting guidelines using CARE Guidelines: https://www.care-statement.org

Authors are encouraged to use the CARE Checklist prior to submission: https://www.care-statement.org/checklist

Acute & Specialty Care Case Report. This Department features case presentations reflecting either acute or specialty care. The anonymity of patients presented should be maintained and permission obtained from the patient and or family if the patient is not de-identified. IRB approval for the presentation of case reports may be required and is the author's responsibility. Any author interested in sending a query should contact the corresponding editor at maddenma@rwjms.rutgers.edu. See the suggested template for case report preparation. The suggested word count is 3000.

Reporting guidelines using CARE Guidelines: https://www.care-statement.org

Authors are encouraged to use the CARE Checklist prior to submission: https://www.care-statement.org/checklist

Health Policy & Professional Issues. *Health Policy*Current and compelling state, national health and international policy issues impacting children and their families are published. The suggested word count for this department is 1500 words.

*Professional Issues*This Department features articles about professional practice, role issues, and leadership topics of interest to pediatric advanced practice nurses.The suggested word count is 2500 words. The corresponding editor for Health Policy and Professional Issues is efrybowers@usfca.edu.

Research Methods. This feature seeks to provide new and innovative research methodologies for pediatric investigations to inform the design, implementation, dissemination, and evaluation of research-based care by PNPs. Analysis of methodologies to advance the science and evidence-base for pediatric and family centered research studies are encouraged. Send queries to the corresponding editor at rspratling@gsu.edu. The suggested word count is 2000 words.

CONTINUING EDUCATION MANUSCRIPT PREPARATION

Continuing education. Manuscripts on non-pharmacologic clinical topics that highlight changes in clinical practice based on the latest available research studies supporting or refuting current practices. This column is a CE offering (see CE instructions below). Queries for this Department can be sent to the associate editor at anne.derouin@duke.edu.

Pharmacology CE. Manuscripts on pharmacologic and psychopharmacology management of pediatric, adolescent, and young adult illnesses are published. Analysis of the pharmacokinetics, pharmacodynamics and pharmacogenomics related to specific medical diagnoses, prescribing practices, drug safety, drug monitoring, and side effects are published. This column is a CE offering (see CE instructions below). Queries for this Department can be sent to the corresponding editor at TWoo@stmartin.edu.

Continuing Education Article Author Information

Manuscripts submitted may offer varying amounts of CE credit. To be considered for CE, a manuscript must include:

1. Demonstration of the prospective author's expertise in the subject matter through experience, education, or both.

2. List 3-5 objectives, using action verbs that require readers to demonstrate their understanding of the topic (e.g., Explain the pathophysiology of...Recommend an appropriate approach...Discuss important considerations...). Use of Bloom's Revised taxonomy is recommended to develop each of the objectives and relevant questions.

3. A researched, referenced manuscript of approximately 6000 words (including objectives, tables, and posttest questions/answers). The text must provide current, advanced, testable information on clinical or professional topics.

4. Ten multiple-choice questions with 4 responses each or true/false items with the correct answers indicated. (See "Tips for Writing Test Questions.") Multiple choice questions with the correct answer of "all of the above" are not acceptable.

5. A curriculum vitae for each author should be sent

to lnelsen@napnap.org upon acceptance.

6. A faculty declaration form for NAPNAP must be completed by all authors. This form can be accessed at http://www.jpedhc.org. Completed forms should be returned to lnelsen@napnap.org.

Tips for Writing Test Questions

Questions should measure mastery of objectives and article content. Ideally, the majority of questions should be designed for the reader to apply knowledge learned from reading the article as opposed to simple recall of information.

1. Be sure the order of questions matches the sequence of information in the article. For example, question #1 should correspond to the information that appears in the article first.

2. After you have finished writing the test, be certain that the test includes questions that relate to each objective.

3. Make questions multiple choice with possible options labeled "a," "b," "c," "d."

4. Be certain that the 3 incorrect options are not plausible.

5. Use the same terminology in the test as in the narrative. (For example, if the narrative refers only to "hypertension," use "hypertension," not "high blood pressure," in the test.)

6. Make sure the correct option is derived directly from the narrative and clearly defensible as the best answer.

7. Avoid using words in the correct option that are also found in the stem (the first part of the question). Doing so provides "clues" to the correct answer.

8. Make sure that the options are not mutually exclusive. For example, if option "a" reads, "Slows the heart rate," and option "b" reads, "Increases the heart rate," these 2 options are mutually exclusive. The test taker can be reasonably certain that "c" and "d" are extraneous, and that either "a" or "b" is the correct answer.

9. Be sure that 1 or more of the options are not included in another option. For example, if option "a" reads, "Affects the heart rate," and option "b"

reads, "Slows the heart rate," option "b" is actually included in option "a." Thus, if "b" is a correct response, "a" is also.

10. Include an answer key. The editor reserves the right to edit questions submitted for purposes of clarity and accuracy. The editors acknowledge the challenge of constructing a posttest that is accurate and clear.

Continuing Education Approval Procedure

1. The number of contact hours is assigned by the Education department of NAPNAP.

2. One member of the CE Committee not associated with the *JPHC*, who matches the profile of the average *JPHC* reader, is asked to review the article. An average of the time it takes them to read the article and complete the posttest will determine the number of contact hour(s) assigned. One contact hour equals 60 minutes.

3. CE Articles are approved for 1 year. At the discretion of the Education department, the approval time may be shortened depending on the stability of the article content.

For more information regarding development of learning objectives and posttest questions, please contact Laura Nelsen at lnelsen@napnap.org.

Peer review

This journal operates a double anonymized review process. All contributions will be initially assessed by the editor for suitability for the journal. Papers deemed suitable are then typically sent to a minimum of two independent expert reviewers to assess the scientific quality of the paper. The Editor is responsible for the final decision regarding acceptance or rejection of articles. The Editor's decision is final. Editors are not involved in decisions about papers which they have written themselves or have been written by family members or colleagues or which relate to products or services in which the editor has an interest. Any such submission is subject to all of the journal's usual procedures, with peer review handled independently of the relevant editor and their research groups. More information on types of peer review.

Double anonymized review

This journal uses double anonymized review, which means the identities of the authors are concealed from the reviewers, and vice versa. More information is available on our website. To facilitate this, please include the following separately:

Title page (with author details): This should include the title, authors' names, affiliations, acknowledgements and any Declaration of Interest statement, and a complete address for the corresponding author including an e-mail address.

Anonymized manuscript (no author details): The main body of the paper (including the references, figures, tables and any acknowledgements) should not include any identifying information, such as the authors' names or affiliations.

Use of word processing software

It is important that the file be saved in the native format of the word processor used. The text should be in single-column format. Keep the layout of the text as simple as possible. Most formatting codes will be removed and replaced on processing the article. In particular, do not use the word processor's options to justify text or to hyphenate words. However, do use bold face, italics, subscripts, superscripts etc. When preparing tables, if you are using a table grid, use only one grid for each individual table and not a grid for each row. If no grid is used, use tabs, not spaces, to align columns. The electronic text should be prepared in a way

very similar to that of conventional manuscripts (see also the Guide to Publishing with Elsevier). Note that source files of figures, tables and text graphics will be required whether or not you embed your figures in the text. See also the section on Electronic artwork.
To avoid unnecessary errors you are strongly advised to use the 'spell-check' and 'grammar-check' functions of your word processor.

Highlights

Highlights are optional yet highly encouraged for this journal, as they increase the discoverability of your article via search engines. They consist of a short collection of bullet points that capture the novel results of your research as well as new methods that were used during the study (if any). Please have a look at the example Highlights.

Highlights should be submitted in a separate editable file in the online submission system. Please use 'Highlights' in the file name and include 3 to 5 bullet points (maximum 85 characters, including spaces, per bullet point).

Units

Follow internationally accepted rules and conventions: use the international system of units (SI). If other units are mentioned, please give their equivalent in SI.

ILLUSTRATION SUBMISSION

Figures must be submitted in electronic format. Images should be provided in EPS or TIFF format per the instructions for online submission at https://www.editorialmanager.com/ymph. Illustrations should be numbered in the order of their mention in the text. Please refer to the Author Artwork Instructions link at the Journal's online submission system (https://www.editorialmanager.com/ymph) for additional information about artwork.

The legends should be typed double-spaced on a separate document and numbered to correspond with the figures. If a figure has been previously published, the legend must give full credit to the original source and permission obtained. Please send the permission and direct any questions to c.conway@elsevier.com.

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Please note: Because of technical complications which can arise by converting color figures to 'gray scale' (for the printed version should you not opt for color in print) please submit in addition usable black and white versions of all the color illustrations.

Figure captions

Ensure that each illustration has a caption. Supply captions separately, not attached to the figure. A caption should comprise a brief title (**not** on the figure itself) and a description of the illustration. Keep text in the illustrations themselves to a minimum but explain all symbols and abbreviations used.

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Each table should be submitted as a separate file. Please ensure each table file is an editable Word document. They should be numbered according to their mention in the text. A concise title describing the table's content should be supplied for each table. All footnotes should appear immediately below the table, and all abbreviations not used in the text should be defined in a footnote. If a table or any data therein have been previously published, a footnote must give full credit to the original source with permission obtained. Please send the permission and direct any questions to c.conway@elsevier.com.

REFERENCES

Use the reference style of the Publication Manual of the American Psychological Association (7th ed., 2020). The reference list should appear on a separate page at the end of the text. Only references cited in the text should appear in this list.

Please know that Google Scholar does not vet articles that are included in searches. Thus, a predatory journal article may be included in your Google Scholar search, and you may have a predatory article in your submitted work. It is your responsibility to check all references to be certain they are valid and have a DOI number prior to submission.

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Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

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As a minimum, the full URL should be given. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

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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), year, dataset title, data repository, version (where available), and global persistent identifier. Add [dataset] immediately before the reference so we can properly identify it as a data reference. This identifier will not appear in your published article.

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Where a preprint has subsequently become available as a peer-reviewed publication, the formal publication should be used as the reference. If there are preprints that are central to your work or that cover crucial developments in the topic, but are not yet formally published, these may be referenced. Preprints should be clearly marked as such, for example by including the word preprint, or the name of the preprint server, as part of the reference. The preprint DOI should also be provided.

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List: references should be arranged first alphabetically and then further sorted chronologically if necessary. More than one reference from the same author(s) in the same year must be identified by the letters 'a', 'b', 'c', etc., placed after the year of publication.

Examples:

Reference to a journal publication:

Burka, S. D., Van Cleve, S. N., Shafer, S., & Barkin, J. L. (2014). At integration of pediatric mental health care: an evidence-based workshop for primary care providers. *Journal of Pediatric Health Care, 28*, 23–34. https://doi.org/10.1016/j.pedhc.2012.10.006

Reference to a chapter in an edited book:

Mettam, G. R., & Adams, L. B. (2009). How to prepare an electronic version of your article. In B. S. Jones, & R. Z. Smith (Eds.), *Introduction to the* *electronic age* (pp. 281–304). New York: E-Publishing Inc.

Reference to a dataset:

[dataset] Oguro, M., Imahiro, S., Saito, S., & Nakashizuka, T. (2015). *Mortality data for Japanese oak wilt disease and surrounding forest composition* [Mendeley Data, v1]. Retrieved from https://doi.org/10.17632/xwj98nb39r.1

Journal abbreviations source

Journal names should be abbreviated according to the list of title word abbreviations: http://www.issn.org/2-22661-LTWA-online.php.

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SUPPLEMENTARY MATERIALS

Supplementary data (i.e., multimedia files, additional images/datasets, etc.) to accompany your manuscript can be submitted. If the manuscript is accepted, such file(s) may appear with the online version of the article and the availability of the online file(s) will be noted in the printed version of the *JPHC*.

Research data

This journal encourages and enables you to share data that supports your research publication where appropriate, and enables you to interlink the data with your published articles. Research data refers to the results of observations or experimentation that validate research findings, which may also include software, code, models, algorithms, protocols, methods and other useful materials related to the project.

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AUTHOR CONTRIBUTIONS

E. A. D. and E. S. conceptualization; E. A. D., H. L., and E. S. resources; E. A. D., Y. L., Z. S., H. L., and E. S. data curation; E. A. D. formal analysis; E. A. D., Y. L., and E. S. supervision; E. A. D. and E. S. funding acquisition; E. A. D. validation; E. A. D. and Z. S. investigation; E. A. D. visualization; E. A.

D. and Y. L. methodology; E. A. D. writing-original draft; E. A. D. and E. S. project administration; E. A. D., Y. L., and E. S. writing-review and editing. For any further information please visit our customer support site at https://service.elsevier.com.

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Appendix 2: Quality Criteria of Included Studies

CASP Quality Appraisal Checklist for Qualitative Studies (CASP, 2018)

Author	Q1.	Q2	Q3.	Q4.	Q5.	Q6.	Q7.	Q8.	Q9.	Q10.
Carlisle et al. (2022)	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Lerrett et al. (2016)	Yes	Yes	Can't tell	Yes	Yes	No	No	Can't tell	Yes	Yes
Waite et al. (2022)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Almoajil et al. (2022)	Yes	Yes	Can't tell	Yes	Yes	No	Yes	Yes	Yes	Yes
Papiez et al. (2021)	Yes	Yes	Yes	Yes	Yes	Can't tell	Yes.	Yes	Yes	Yes
Boss et al. (2017)	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Heath et al. (2016)	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Lecouturier et al. (2015)	Yes	Yes	Cant tell	Yes	Yes	No	Yes	Yes	Can't tell	Yes
Samata et al. (2022)	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Smith et al. (2013b)	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	No
Timmermans et al (2018).	Yes	Yes	Yes	No	Yes	No	Can't tell.	Yes	Yes	Yes
Sjoberg et al. (2015).	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes

CASP Quality Appraisal Checklist for Cohort Studies (CASP, 2018)

Author	Q1	Q2	Q3	Q4	Q5 A & B	Q6 A & B	Q7	Q8	Q9	Q10	Q11	Q12
Links et al., (2020)	Yes	Yes	Yes	Yes	No Can't tell	No Yes	Yes	Yes	Yes	No	Yes	Yes

JBI Critical Appraisal Checklist for Case Reports (JBI, 2020)

Author	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Rating

Loeff & Shakhsheer (2021)	Yes	Yes	Yes	Yes	Yes	N/A	No/Unclear	Yes	Low

JBI Critical Appraisal Checklist for Systematic Reviews and Research Syntheses (JBI, 2020)

Author	**Q1**	**Q2**	**Q3**	**Q4**	**Q5**	**Q6**	**Q7**	**Q8**	**Q9**	**Q10**	**Q11**	**Rating**
Atsaidis et al., (2022).	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Unclear	N/A	Yes	No	Medium

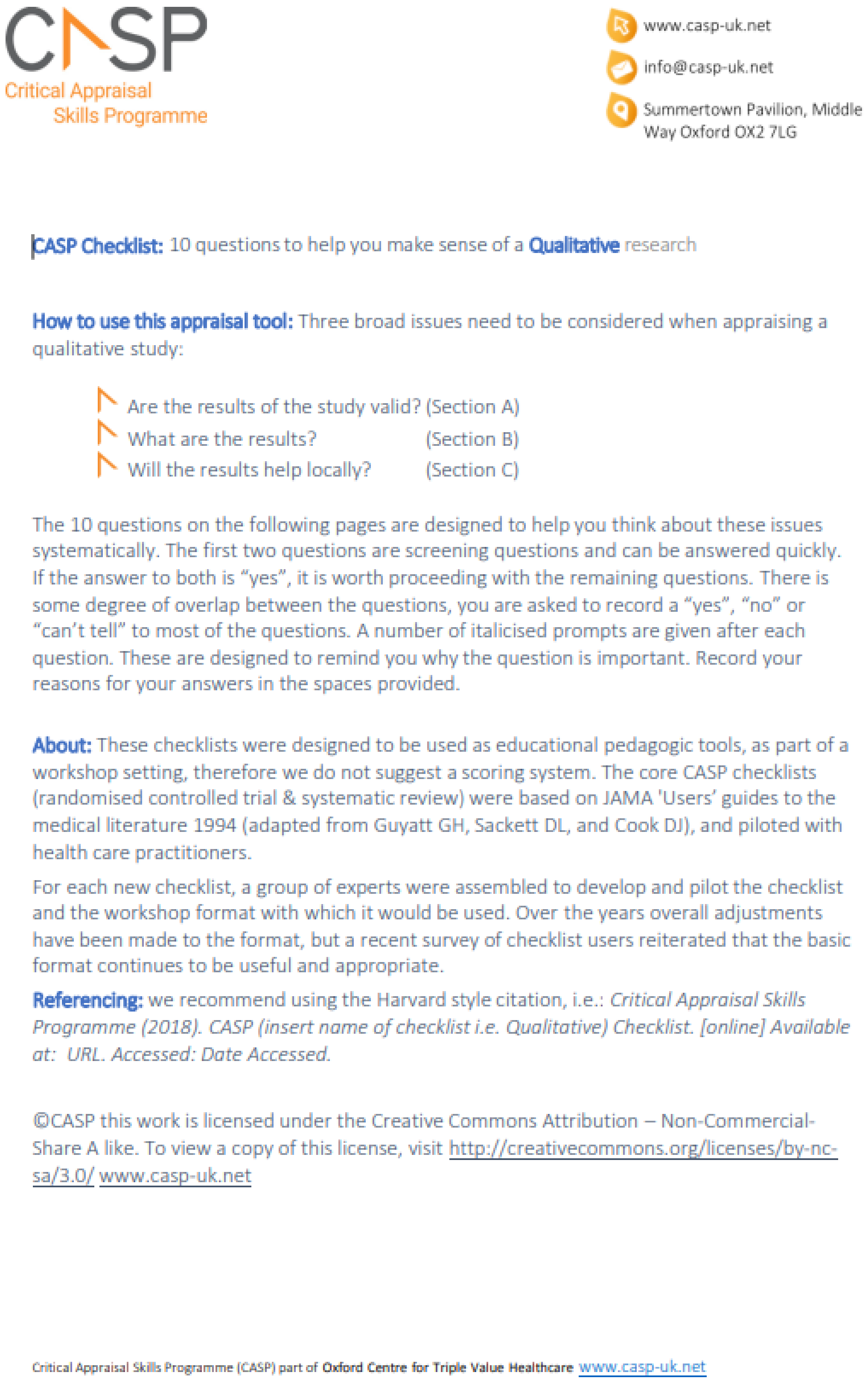
Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies – NHLBI, NIH (2013)

Author	**Q1**	**Q2**	**Q3**	**Q4**	**Q5**	**Q6**	**Q7**	**Q8**	**Q9**	**Q10**	**Q11**	**Q12**	**Q13**	**Q14**	**Rating**
Leu et al., (2021)	Y	N	N	CD	N	Y	N/A	N/A	N	N/A	Y	N/A	N/A	Y	Poor

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

Author and Year	**Q1**	**Q2**	**Q3**	**Q4**	**Q5**
Smith et al., (2013c)	Yes	No	No	Yes – no divergence.	Yes

Appendix 3: CASP Checlist for appraising Qualitative Studies



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CASP Checklist: 10 questions to help you make sense of a **Qualitative** research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

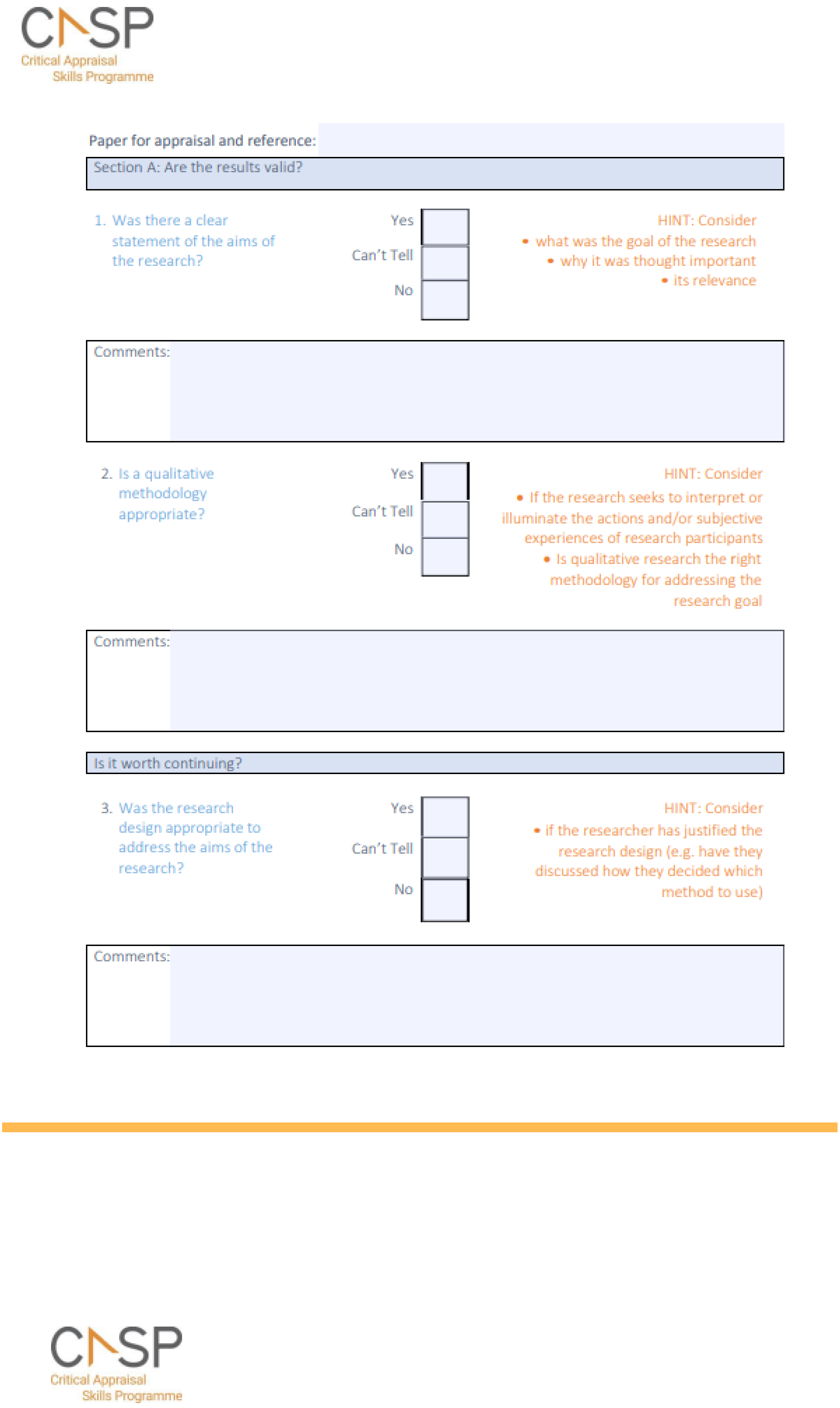
- Are the results of the study valid? (Section A)
- What are the results? (Section B)
- Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is “yes”, it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a “yes”, “no” or “can’t tell” to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users’ guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*



Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- what was the goal of the research
- why it was thought important
- its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
- Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

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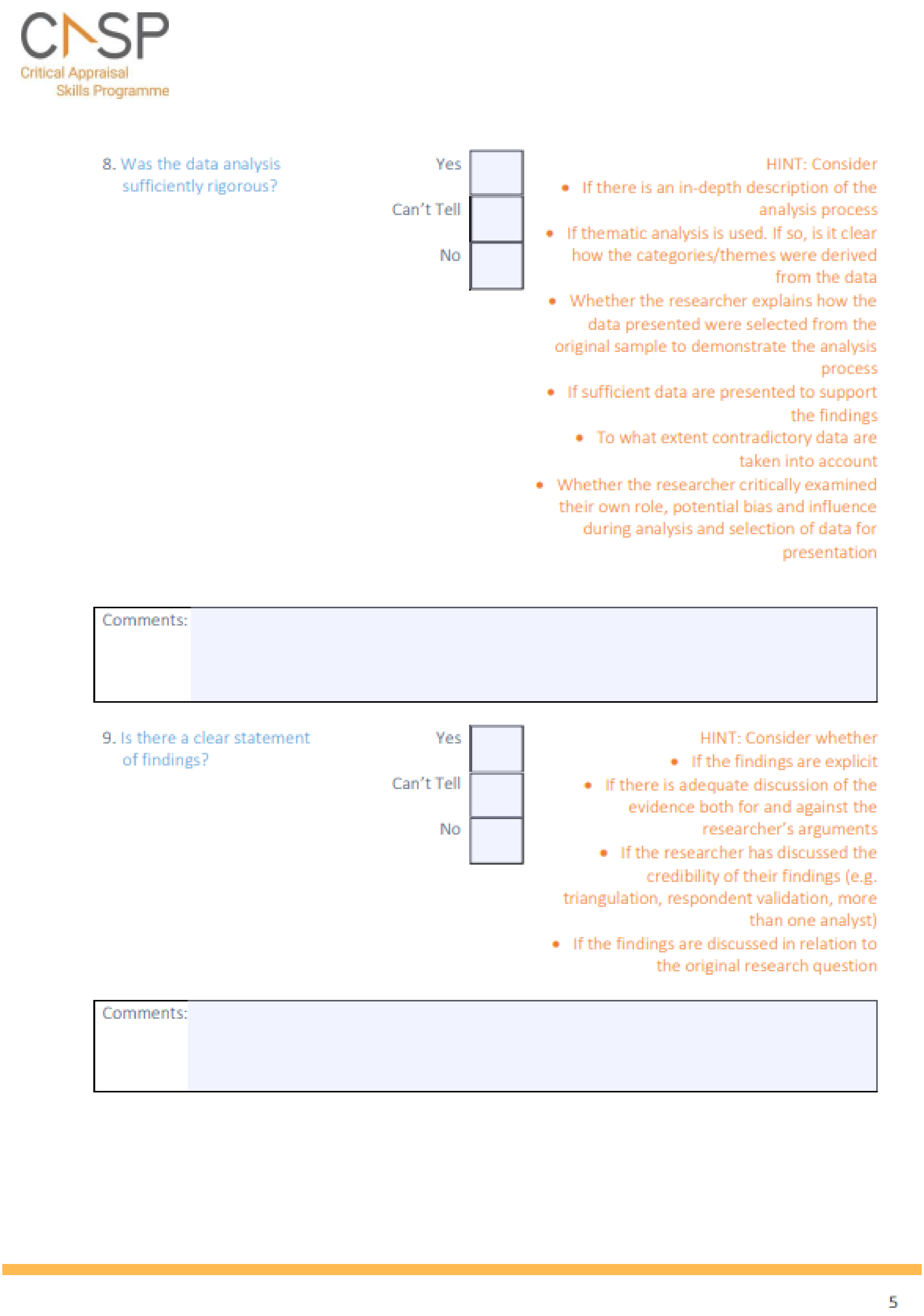

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8. Was the data analysis sufficiently rigorous?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
- To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

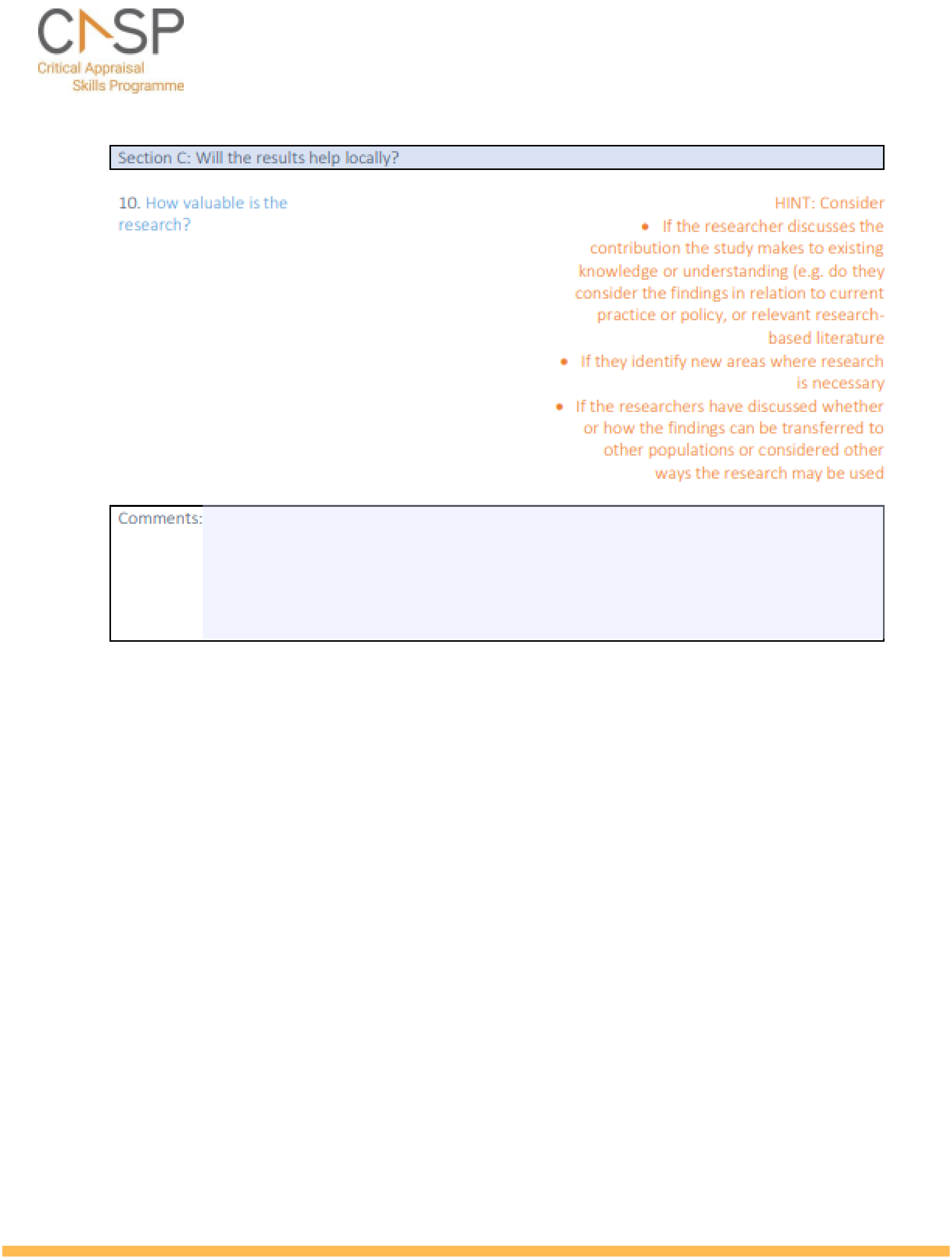
9. Is there a clear statement of findings?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider whether
- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

5



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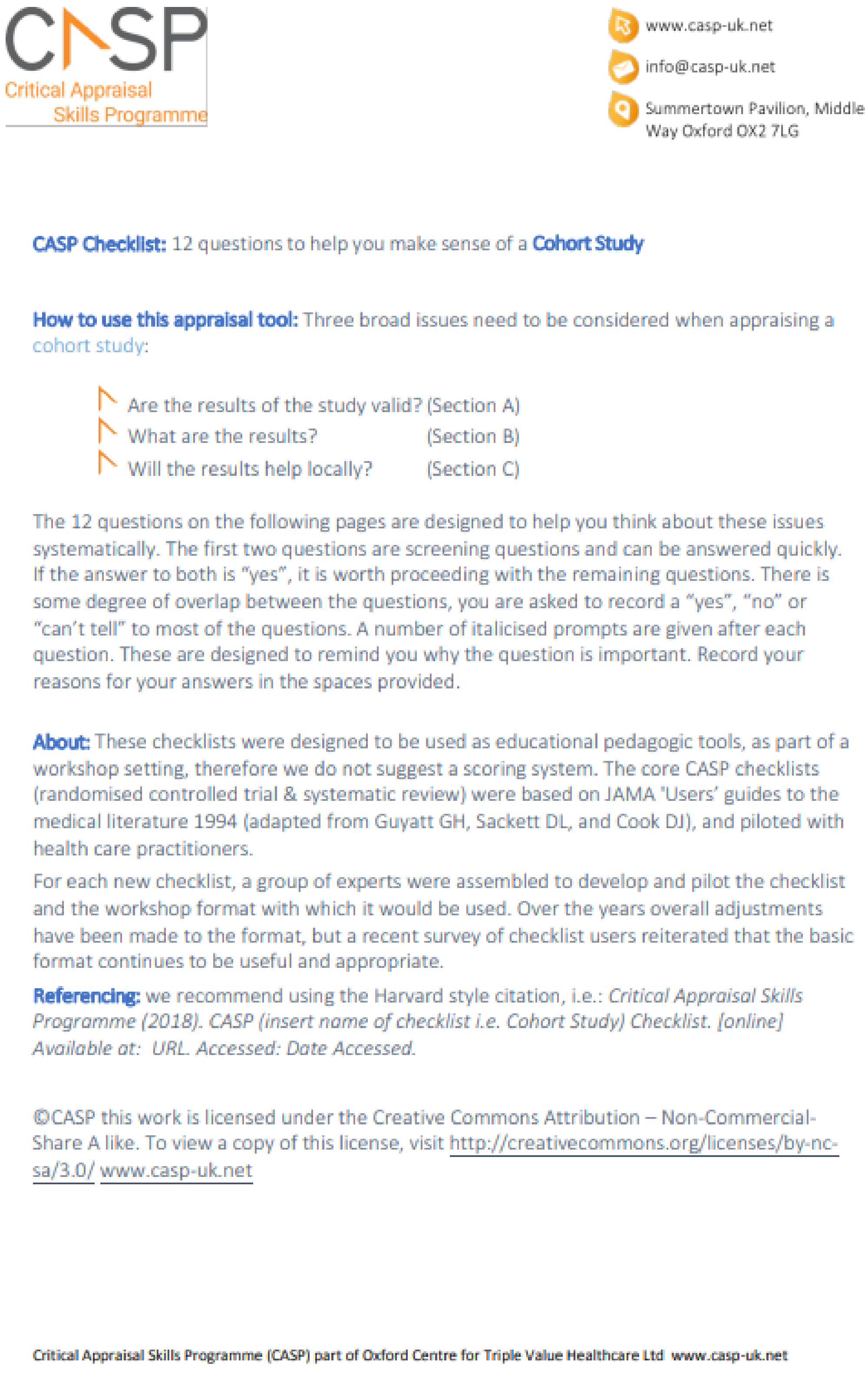
Section C: Will the results help locally?

10. How valuable is the research?	HINT: Consider • If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature • If they identify new areas where research is necessary • If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Appendix 4: CASP Checklist for Appraising Cohort Studies



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CASP Checklist: 12 questions to help you make sense of a **Cohort Study**

How to use this appraisal tool: Three broad issues need to be considered when appraising a cohort study:

- Are the results of the study valid? (Section A)
- What are the results? (Section B)
- Will the results help locally? (Section C)

The 12 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is "yes", it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a "yes", "no" or "can't tell" to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

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Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Cohort Study) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

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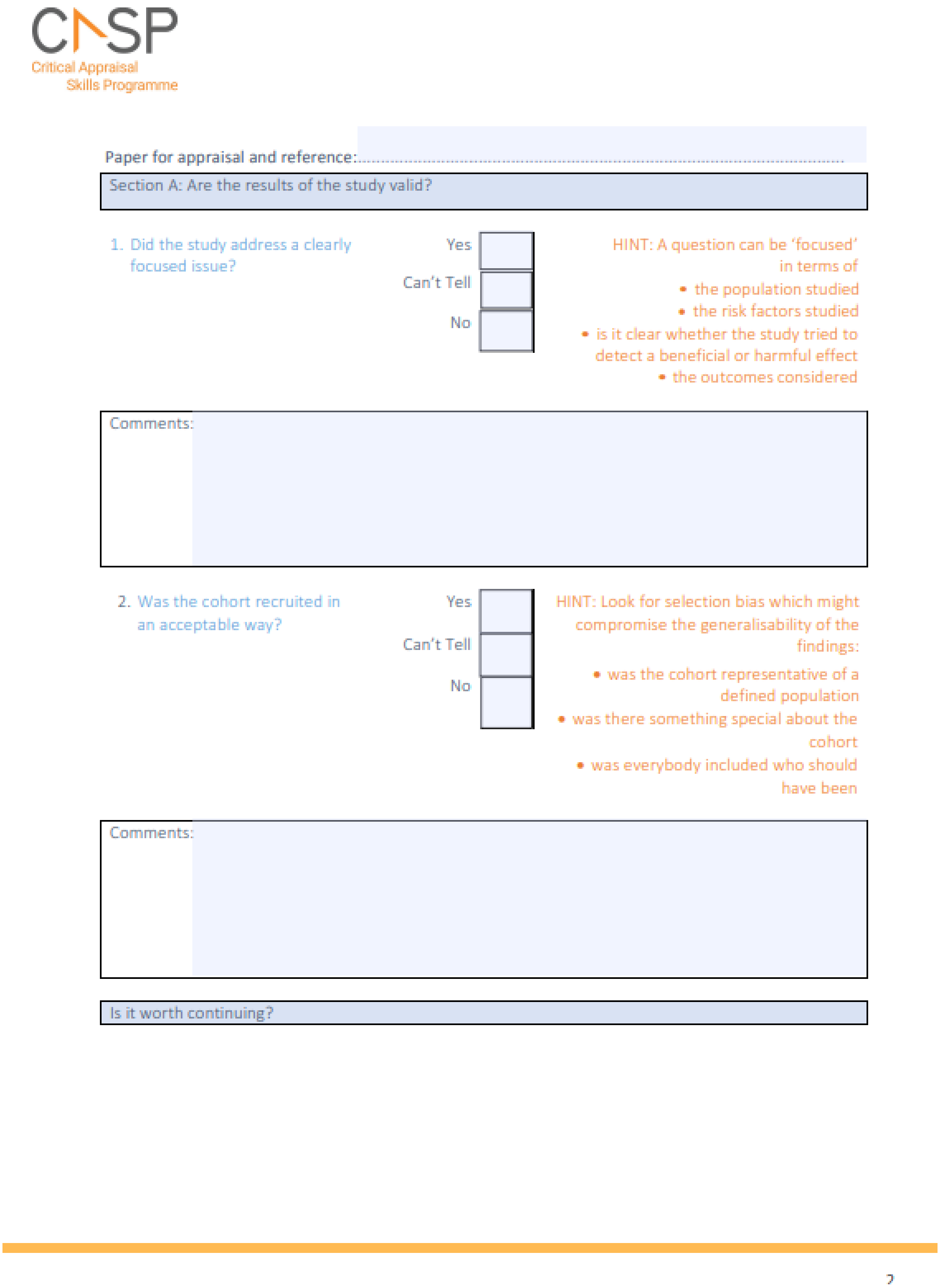

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Paper for appraisal and reference:...

Section A: Are the results of the study valid?

1. Did the study address a clearly focused issue?	Yes ☐ Can't Tell ☐ No ☐	HINT: A question can be 'focused' in terms of • the population studied • the risk factors studied • is it clear whether the study tried to detect a beneficial or harmful effect • the outcomes considered

Comments:

2. Was the cohort recruited in an acceptable way?	Yes ☐ Can't Tell ☐ No ☐	HINT: Look for selection bias which might compromise the generalisability of the findings: • was the cohort representative of a defined population • was there something special about the cohort • was everybody included who should have been

Comments:

Is it worth continuing?

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3. Was the exposure accurately measured to minimise bias?

Yes ☐
Can't Tell ☐
No ☐

HINT: Look for measurement or classification bias:

- did they use subjective or objective measurements
- do the measurements truly reflect what you want them to (have they been validated)
- were all the subjects classified into exposure groups using the same procedure

Comments:

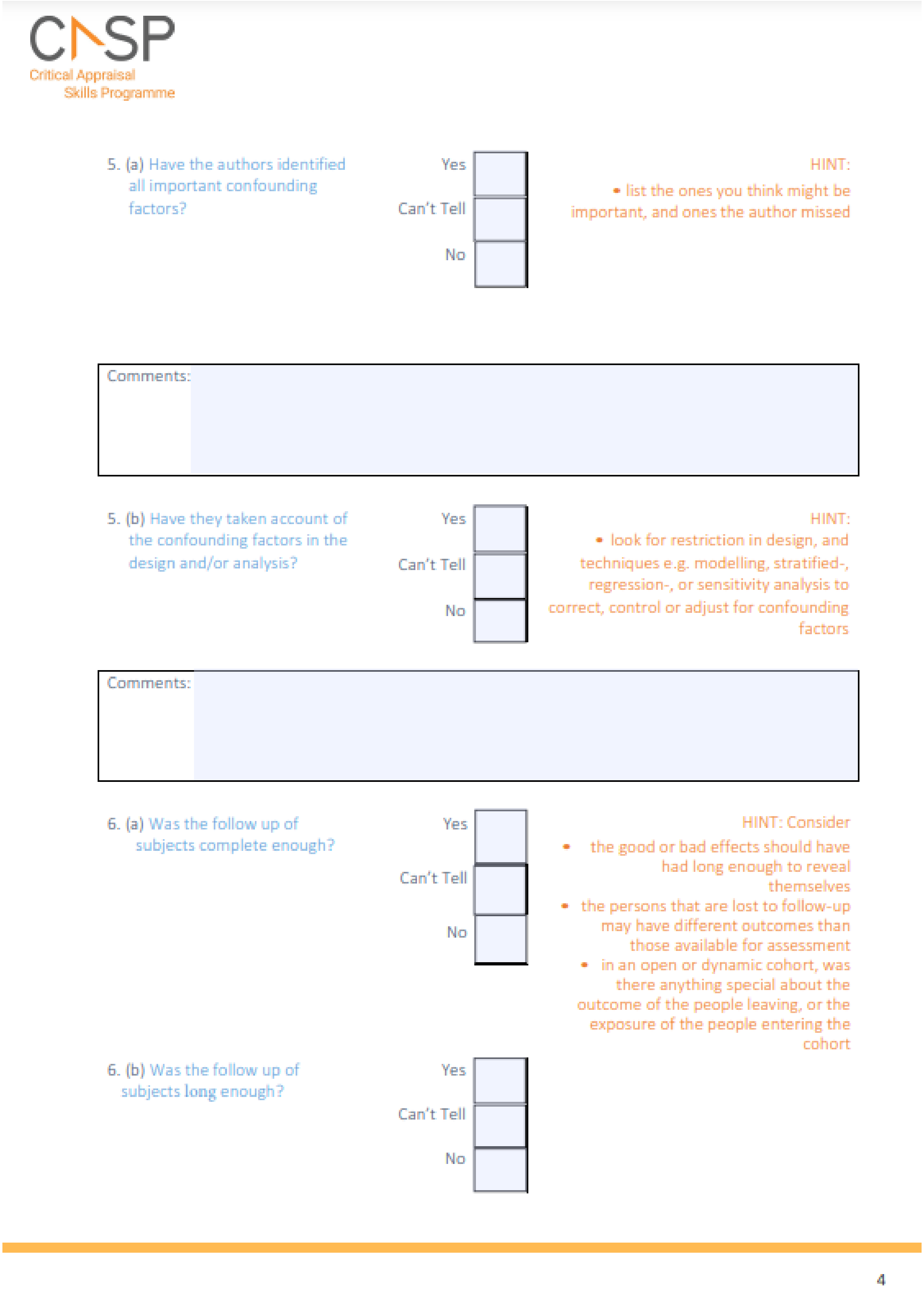
4. Was the outcome accurately measured to minimise bias?

Yes ☐
Can't Tell ☐
No ☐

HINT: Look for measurement or classification bias:

- did they use subjective or objective measurements
- do the measurements truly reflect what you want them to (have they been validated)
- has a reliable system been established for detecting all the cases (for measuring disease occurrence)
- were the measurement methods similar in the different groups
- were the subjects and/or the outcome assessor blinded to exposure (does this matter)

Comments:



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5. (a) Have the authors identified all important confounding factors?

Yes ☐
Can't Tell ☐
No ☐

HINT:
- list the ones you think might be important, and ones the author missed

Comments:

5. (b) Have they taken account of the confounding factors in the design and/or analysis?

Yes ☐
Can't Tell ☐
No ☐

HINT:
- look for restriction in design, and techniques e.g. modelling, stratified-, regression-, or sensitivity analysis to correct, control or adjust for confounding factors

Comments:

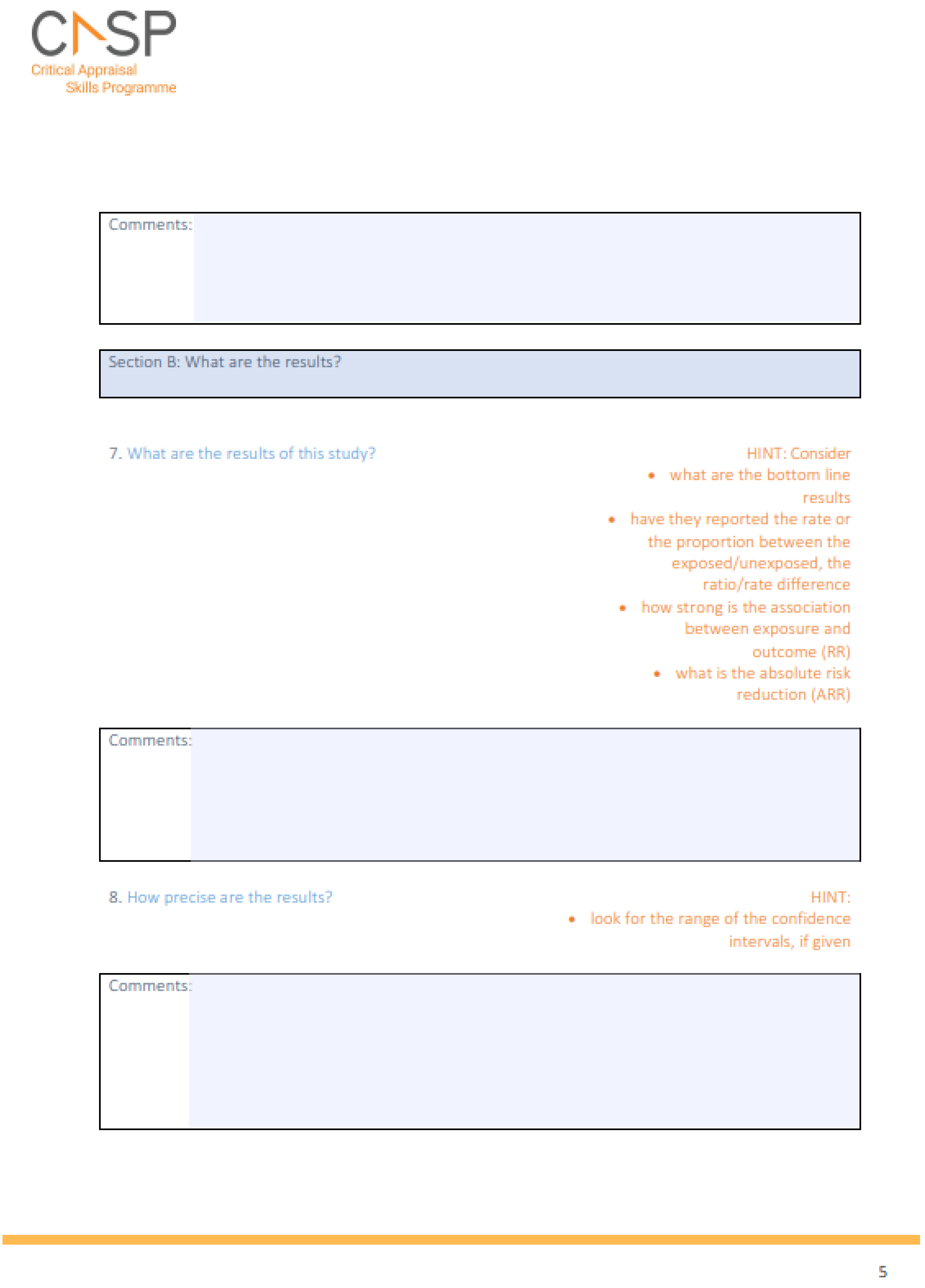
6. (a) Was the follow up of subjects complete enough?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- the good or bad effects should have had long enough to reveal themselves
- the persons that are lost to follow-up may have different outcomes than those available for assessment
- in an open or dynamic cohort, was there anything special about the outcome of the people leaving, or the exposure of the people entering the cohort

6. (b) Was the follow up of subjects long enough?

Yes ☐
Can't Tell ☐
No ☐



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Comments:

Section B: What are the results?

7. What are the results of this study?

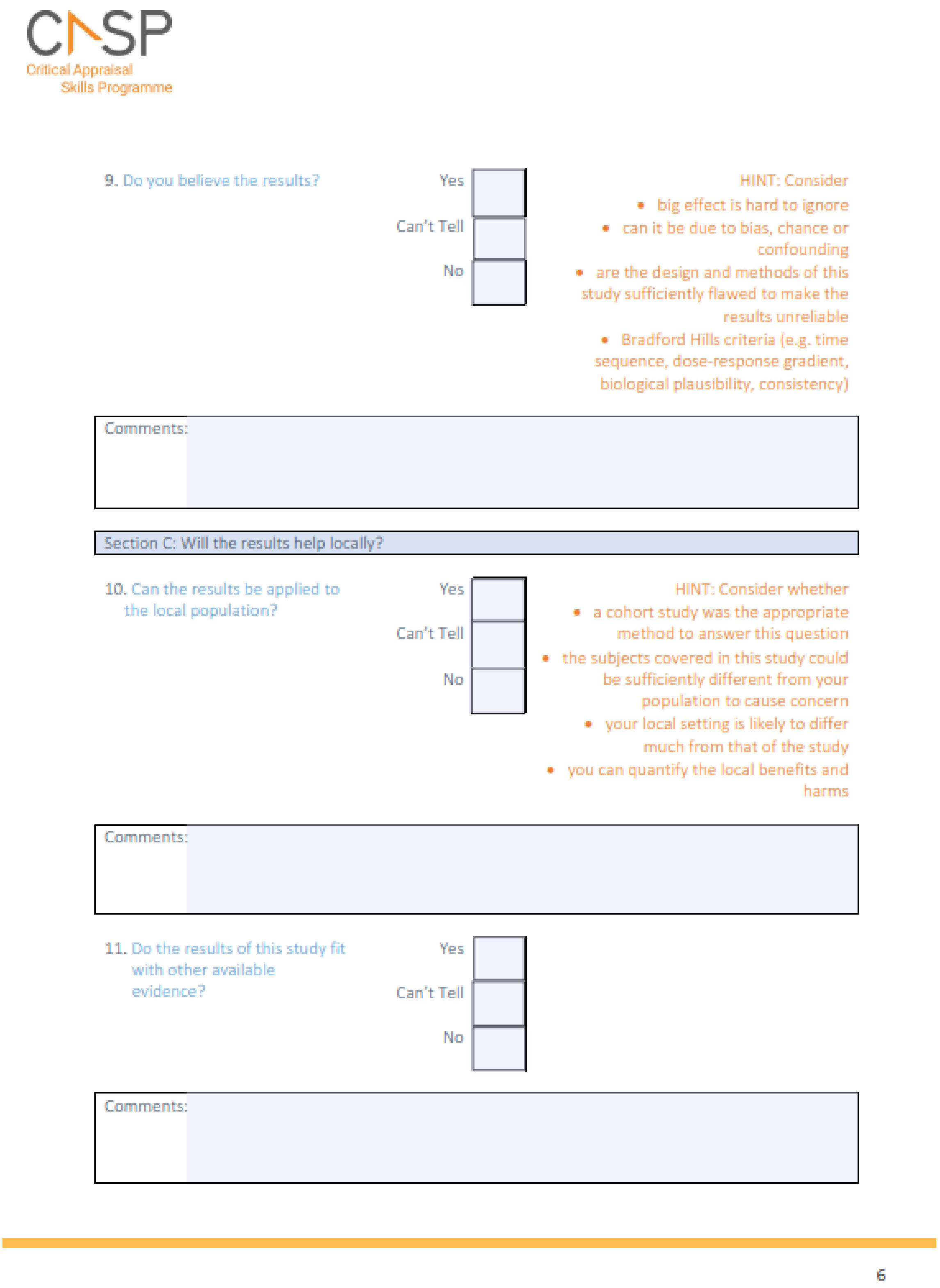
HINT: Consider
- what are the bottom line results
- have they reported the rate or the proportion between the exposed/unexposed, the ratio/rate difference
- how strong is the association between exposure and outcome (RR)
- what is the absolute risk reduction (ARR)

Comments:

8. How precise are the results?

HINT:
- look for the range of the confidence intervals, if given

Comments:



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9. Do you believe the results?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- big effect is hard to ignore
- can it be due to bias, chance or confounding
- are the design and methods of this study sufficiently flawed to make the results unreliable
- Bradford Hills criteria (e.g. time sequence, dose-response gradient, biological plausibility, consistency)

Comments:

Section C: Will the results help locally?

10. Can the results be applied to the local population?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider whether
- a cohort study was the appropriate method to answer this question
- the subjects covered in this study could be sufficiently different from your population to cause concern
- your local setting is likely to differ much from that of the study
- you can quantify the local benefits and harms

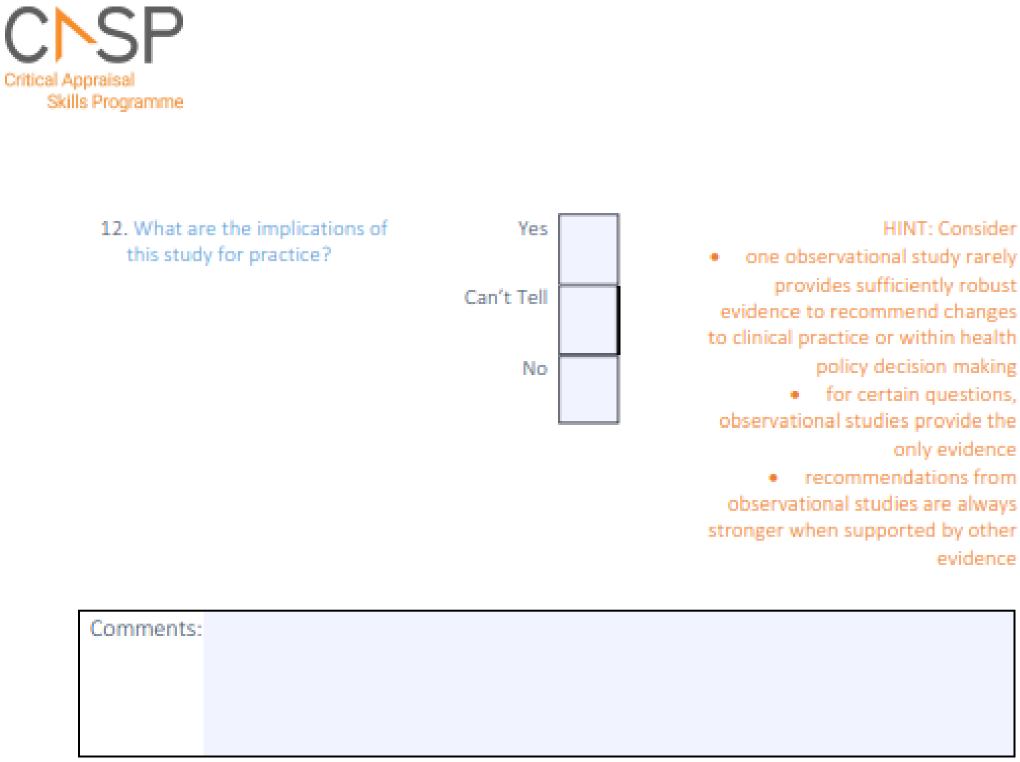
Comments:

11. Do the results of this study fit with other available evidence?

Yes ☐
Can't Tell ☐
No ☐

Comments:

6



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12. What are the implications of this study for practice?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- one observational study rarely provides sufficiently robust evidence to recommend changes to clinical practice or within health policy decision making
- for certain questions, observational studies provide the only evidence
- recommendations from observational studies are always stronger when supported by other evidence

Comments:

Appendix 5: JBI Checklist for Appraising Case Reports

JBI CRITICAL APPRAISAL CHECKLIST FOR CASE REPORTS

Reviewer______________________________________Date_______________________________

Author_______________________________________Year_________ Record Number_________

	Yes	No	Unclear	Not applicable
1. Were patient's demographic characteristics clearly described?	☐	☐	☐	☐
2. Was the patient's history clearly described and presented as a timeline?	☐	☐	☐	☐
3. Was the current clinical condition of the patient on presentation clearly described?	☐	☐	☐	☐
4. Were diagnostic tests or assessment methods and the results clearly described?	☐	☐	☐	☐
5. Was the intervention(s) or treatment procedure(s) clearly described?	☐	☐	☐	☐
6. Was the post-intervention clinical condition clearly described?	☐	☐	☐	☐
7. Were adverse events (harms) or unanticipated events identified and described?	☐	☐	☐	☐
8. Does the case report provide takeaway lessons?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

____
____
____

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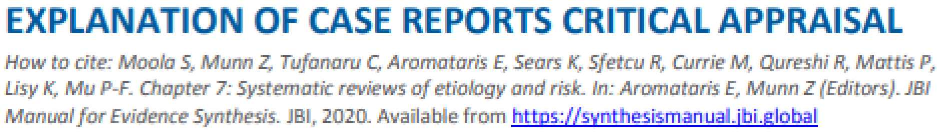
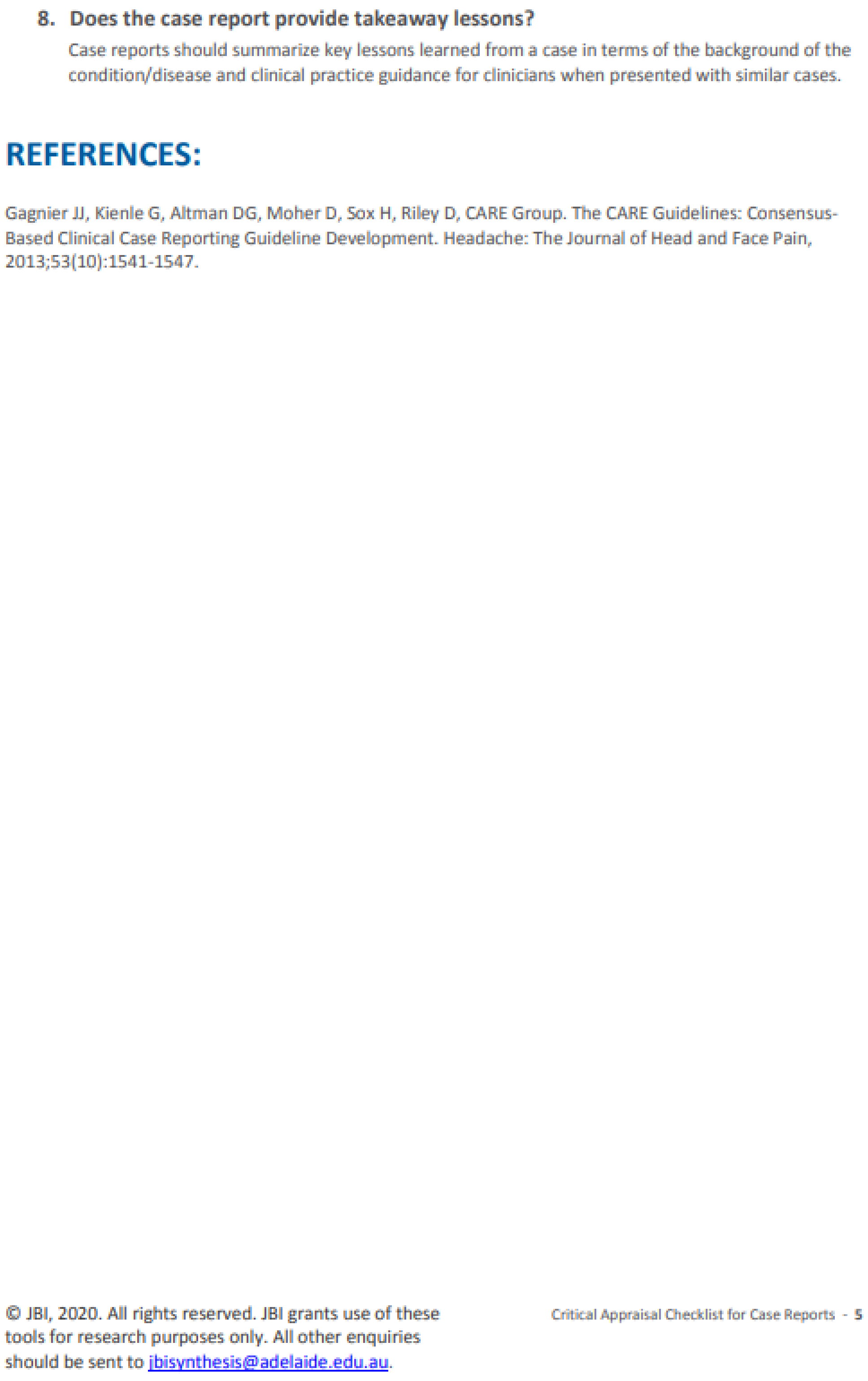
Critical Appraisal Checklist for Case Reports - 3



EXPLANATION OF CASE REPORTS CRITICAL APPRAISAL

How to cite: Moola S, Munn Z, Tufanaru C, Aromataris E, Sears K, Sfetcu R, Currie M, Qureshi R, Mattis P, Lisy K, Mu P-F. Chapter 7: Systematic reviews of etiology and risk. In: Aromataris E, Munn Z (Editors). JBI Manual for Evidence Synthesis. JBI, 2020. Available from https://synthesismanual.jbi.global



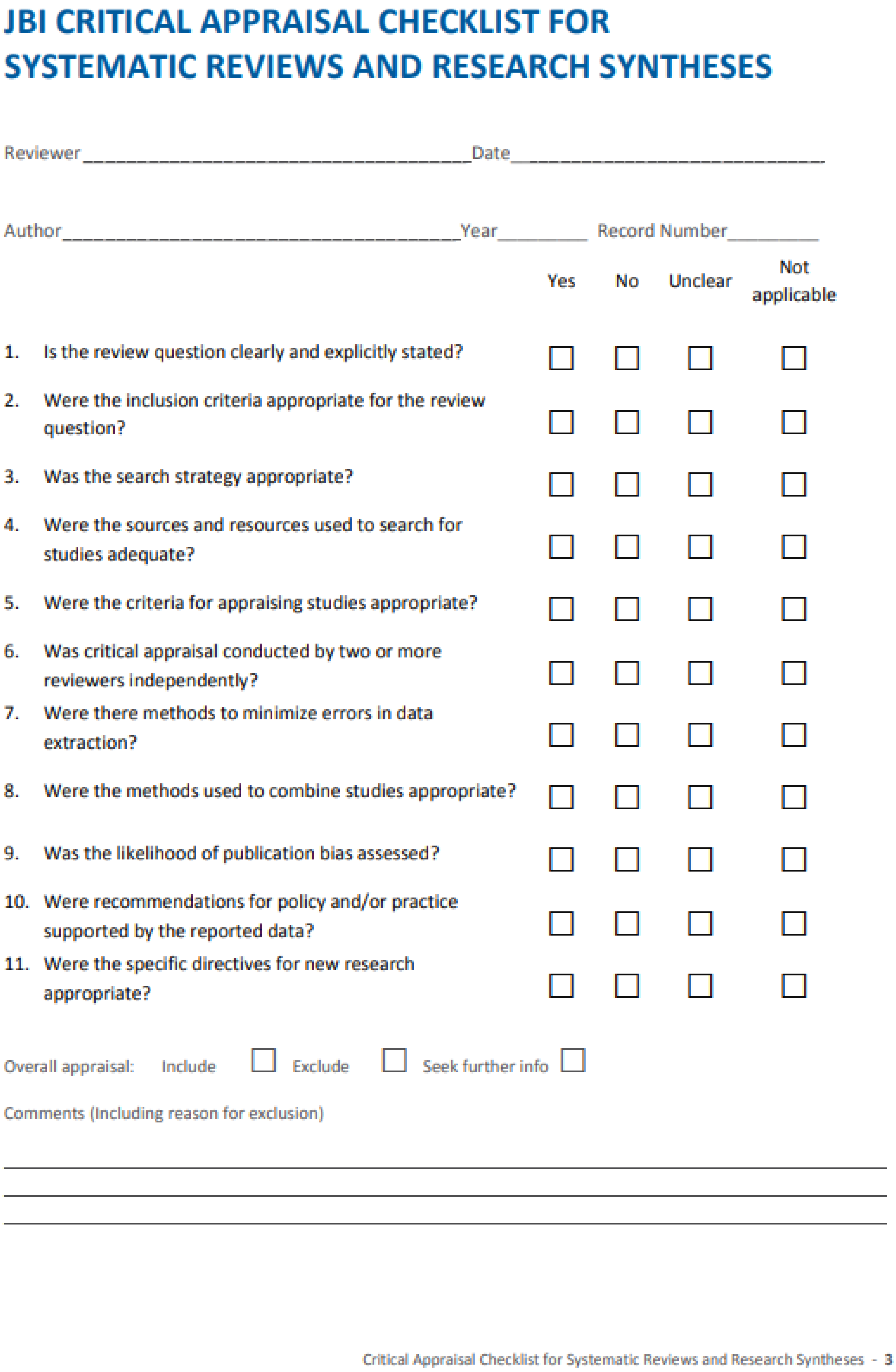
8. **Does the case report provide takeaway lessons?**
   Case reports should summarize key lessons learned from a case in terms of the background of the condition/disease and clinical practice guidance for clinicians when presented with similar cases.

REFERENCES:

Gagnier JJ, Kienle G, Altman DG, Moher D, Sox H, Riley D, CARE Group. The CARE Guidelines: Consensus-Based Clinical Case Reporting Guideline Development. Headache: The Journal of Head and Face Pain, 2013;53(10):1541-1547.

Critical Appraisal Checklist for Case Reports - 5

Appendix 6: JBI Checklist for Systematic Reviews and Research Syntheses



JBI CRITICAL APPRAISAL CHECKLIST FOR SYSTEMATIC REVIEWS AND RESEARCH SYNTHESES

Reviewer______________________________________Date_______________________________

Author_______________________________________Year_________ Record Number_________

	Yes	No	Unclear	Not applicable
1. Is the review question clearly and explicitly stated?	☐	☐	☐	☐
2. Were the inclusion criteria appropriate for the review question?	☐	☐	☐	☐
3. Was the search strategy appropriate?	☐	☐	☐	☐
4. Were the sources and resources used to search for studies adequate?	☐	☐	☐	☐
5. Were the criteria for appraising studies appropriate?	☐	☐	☐	☐
6. Was critical appraisal conducted by two or more reviewers independently?	☐	☐	☐	☐
7. Were there methods to minimize errors in data extraction?	☐	☐	☐	☐
8. Were the methods used to combine studies appropriate?	☐	☐	☐	☐
9. Was the likelihood of publication bias assessed?	☐	☐	☐	☐
10. Were recommendations for policy and/or practice supported by the reported data?	☐	☐	☐	☐
11. Were the specific directives for new research appropriate?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

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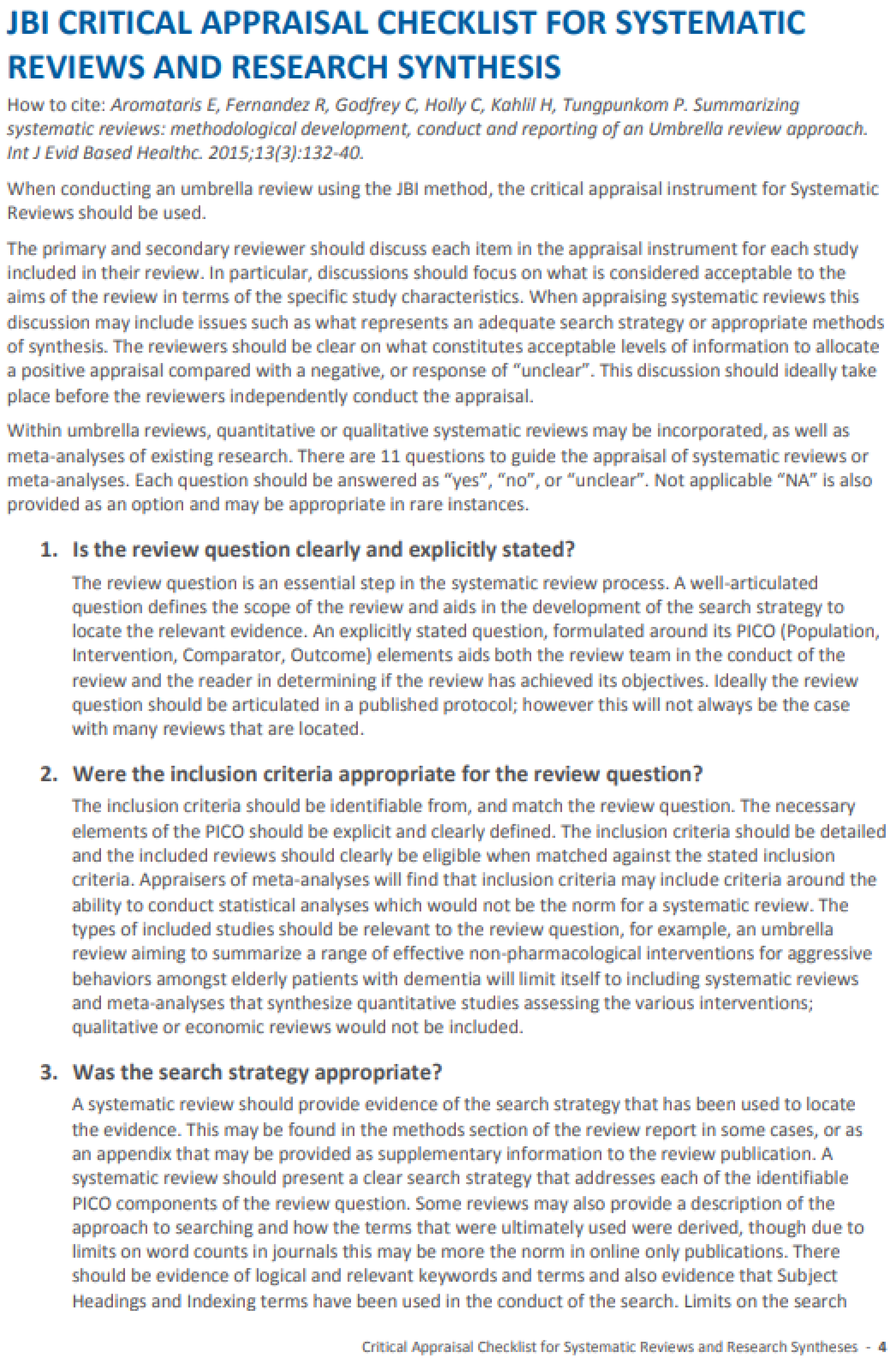
Critical Appraisal Checklist for Systematic Reviews and Research Syntheses - 3



JBI CRITICAL APPRAISAL CHECKLIST FOR SYSTEMATIC REVIEWS AND RESEARCH SYNTHESIS

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When conducting an umbrella review using the JBI method, the critical appraisal instrument for Systematic Reviews should be used.

The primary and secondary reviewer should discuss each item in the appraisal instrument for each study included in their review. In particular, discussions should focus on what is considered acceptable to the aims of the review in terms of the specific study characteristics. When appraising systematic reviews this discussion may include issues such as what represents an adequate search strategy or appropriate methods of synthesis. The reviewers should be clear on what constitutes acceptable levels of information to allocate a positive appraisal compared with a negative, or response of “unclear”. This discussion should ideally take place before the reviewers independently conduct the appraisal.

Within umbrella reviews, quantitative or qualitative systematic reviews may be incorporated, as well as meta-analyses of existing research. There are 11 questions to guide the appraisal of systematic reviews or meta-analyses. Each question should be answered as “yes”, “no”, or “unclear”. Not applicable “NA” is also provided as an option and may be appropriate in rare instances.

1. Is the review question clearly and explicitly stated?

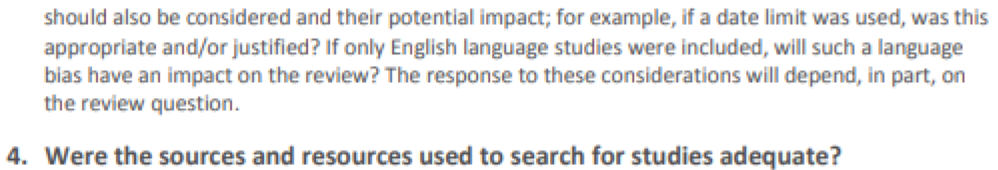
The review question is an essential step in the systematic review process. A well-articulated question defines the scope of the review and aids in the development of the search strategy to locate the relevant evidence. An explicitly stated question, formulated around its PICO (Population, Intervention, Comparator, Outcome) elements aids both the review team in the conduct of the review and the reader in determining if the review has achieved its objectives. Ideally the review question should be articulated in a published protocol; however this will not always be the case with many reviews that are located.

2. Were the inclusion criteria appropriate for the review question?

The inclusion criteria should be identifiable from, and match the review question. The necessary elements of the PICO should be explicit and clearly defined. The inclusion criteria should be detailed and the included reviews should clearly be eligible when matched against the stated inclusion criteria. Appraisers of meta-analyses will find that inclusion criteria may include criteria around the ability to conduct statistical analyses which would not be the norm for a systematic review. The types of included studies should be relevant to the review question, for example, an umbrella review aiming to summarize a range of effective non-pharmacological interventions for aggressive behaviors amongst elderly patients with dementia will limit itself to including systematic reviews and meta-analyses that synthesize quantitative studies assessing the various interventions; qualitative or economic reviews would not be included.

3. Was the search strategy appropriate?

A systematic review should provide evidence of the search strategy that has been used to locate the evidence. This may be found in the methods section of the review report in some cases, or as an appendix that may be provided as supplementary information to the review publication. A systematic review should present a clear search strategy that addresses each of the identifiable PICO components of the review question. Some reviews may also provide a description of the approach to searching and how the terms that were ultimately used were derived, though due to limits on word counts in journals this may be more the norm in online only publications. There should be evidence of logical and relevant keywords and terms and also evidence that Subject Headings and Indexing terms have been used in the conduct of the search. Limits on the search



should also be considered and their potential impact; for example, if a date limit was used, was this appropriate and/or justified? If only English language studies were included, will such a language bias have an impact on the review? The response to these considerations will depend, in part, on the review question.

4. Were the sources and resources used to search for studies adequate?

methods that have been used to synthesize findings congruent with the stated methodology of the review? Is there adequate descriptive and explanatory information to support the final synthesized findings that have been constructed from the findings sourced from the original research?

9. Was the likelihood of publication bias assessed?

As mentioned, a comprehensive search strategy is the best means by which a review author may alleviate the impact of publication bias on the results of the review. Reviews may also present statistical tests such as Egger's test or funnel plots to also assess the potential presence of publication bias and its potential impact on the results of the review. This question will not be applicable to systematic reviews of qualitative evidence.

10. Were recommendations for policy and/or practice supported by the reported data?

Whilst the first nine (9) questions specifically look to identify potential bias in the conduct of a systematic review, the final questions are more indictors of review quality rather than validity. Ideally a review should present recommendations for policy and practice. Where these recommendations are made there should be a clear link to the results of the review. Is there evidence that the strength of the findings and the quality of the research been considered in the formulation of review recommendations?

11. Were the specific directives for new research appropriate?

The systematic review process is recognized for its ability to identify where gaps in the research, or knowledge base, around a particular topic exist. Most systematic review authors will provide some indication, often in the discussion section of the report, of where future research direction should lie. Where evidence is scarce or sample sizes that support overall estimates of effect are small and effect estimates are imprecise, repeating similar research to those identified by the review may be necessary and appropriate. In other instances, the case for new research questions to investigate the topic may be warranted.

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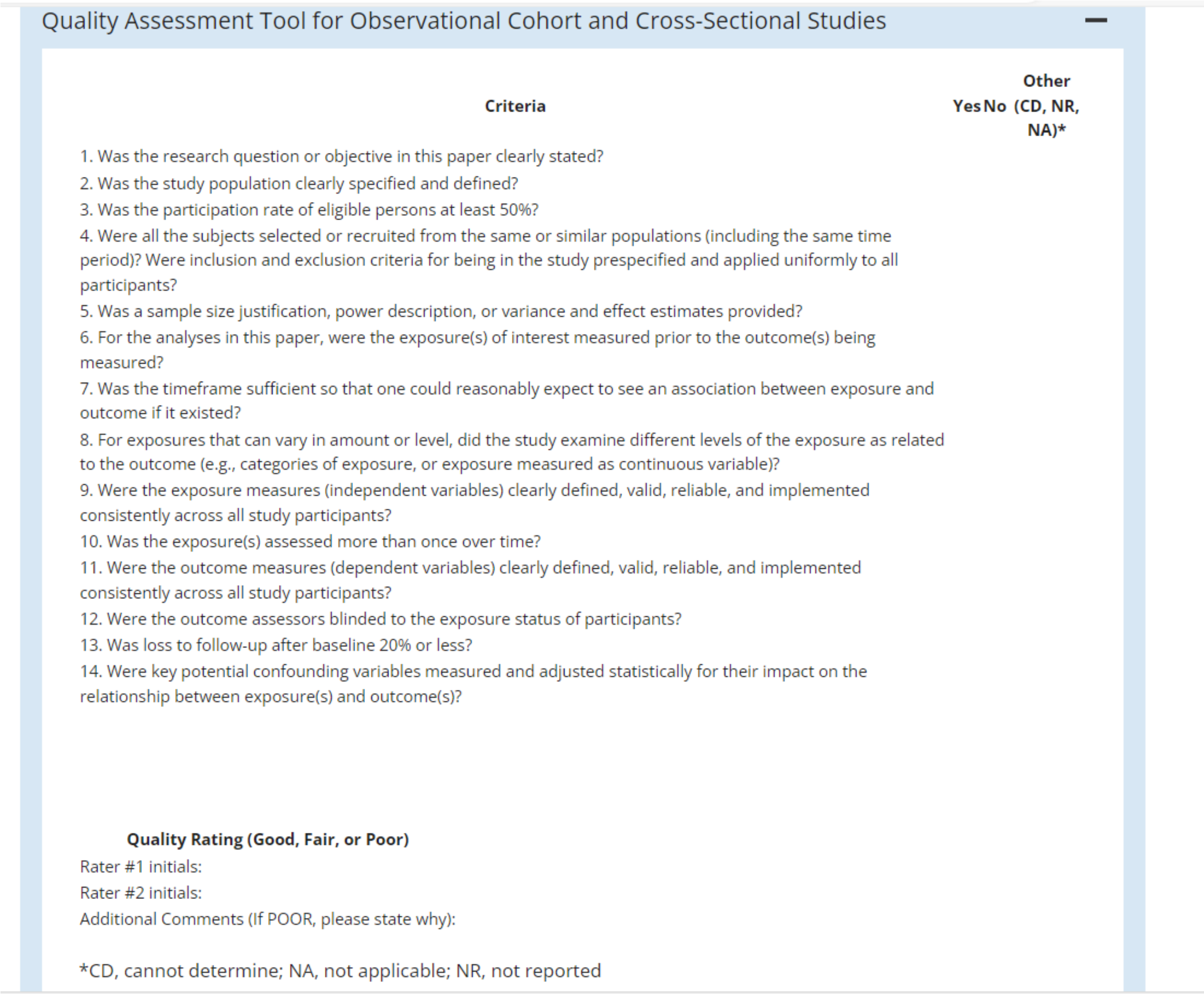
1. Whiting P, Rutjes AWS, Reitsma JB, Bossuyt PMM, Kleijnen J. The development of QUADAS: a tool for the quality assessment of studies of diagnostic accuracy included in systematic reviews. BMC Medical Research Methodology. 2003;3:25 doi:10.1186/1471-2288-3-25.

Appendix 7: NHLBI Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies



Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies

Criteria	Yes	No	Other (CD, NR, NA)*
1. Was the research question or objective in this paper clearly stated?			
2. Was the study population clearly specified and defined?			
3. Was the participation rate of eligible persons at least 50%?			
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study prespecified and applied uniformly to all participants?			
5. Was a sample size justification, power description, or variance and effect estimates provided?			
6. For the analyses in this paper, were the exposure(s) of interest measured prior to the outcome(s) being measured?			
7. Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?			
8. For exposures that can vary in amount or level, did the study examine different levels of the exposure as related to the outcome (e.g., categories of exposure, or exposure measured as continuous variable)?			
9. Were the exposure measures (independent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
10. Was the exposure(s) assessed more than once over time?			
11. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
12. Were the outcome assessors blinded to the exposure status of participants?			
13. Was loss to follow-up after baseline 20% or less?			
14. Were key potential confounding variables measured and adjusted statistically for their impact on the relationship between exposure(s) and outcome(s)?			

Quality Rating (Good, Fair, or Poor)

Rater #1 initials:

Rater #2 initials:

Additional Comments (If POOR, please state why):

*CD, cannot determine; NA, not applicable; NR, not reported

Guidance for Assessing the Quality of Observational Cohort and Cross-Sectional Studies

The guidance document below is organized by question number from the tool for quality assessment of observational cohort and cross-sectional studies.

Question 1. Research question

Did the authors describe their goal in conducting this research? Is it easy to understand what they were looking to find? This issue is important for any scientific paper of any type. Higher quality scientific research explicitly defines a research question.

Questions 2 and 3. Study population

Did the authors describe the group of people from which the study participants were selected or recruited, using demographics, location, and time period? If you were to conduct this study again, would you know who to recruit, from where, and from what time period? Is the cohort population free of the outcomes of interest at the time they were recruited?

An example would be men over 40 years old with type 2 diabetes who began seeking medical care at Phoenix Good Samaritan Hospital between January 1, 1990 and December 31, 1994. In this example, the population is clearly described as: (1) who (men over 40 years old with type 2 diabetes); (2) where (Phoenix Good Samaritan Hospital); and (3) when (between January 1, 1990 and December 31, 1994). Another example is women ages 34 to 59 years of age in 1980 who were in the nursing profession and had no known coronary disease, stroke, cancer, hypercholesterolemia, or diabetes, and were recruited from the 11 most populous States, with contact information obtained from State nursing boards.

In cohort studies, it is crucial that the population at baseline is free of the outcome of interest. For example, the nurses' population above would be an appropriate group in which to study incident coronary disease. This information is usually found either in descriptions of population recruitment, definitions of variables, or inclusion/exclusion criteria.

You may need to look at prior papers on methods in order to make the assessment for this question. Those papers are usually in the reference list.

If fewer than 50% of eligible persons participated in the study, then there is concern that the study population does not adequately represent the target population. This increases the risk of bias.

Question 4. Groups recruited from the same population and uniform eligibility criteria

Were the inclusion and exclusion criteria developed prior to recruitment or selection of the study population? Were the same underlying criteria used for all of the subjects involved? This issue is related to the description of the study population, above, and you may find the information for both of these questions in the same section of the paper.

Most cohort studies begin with the selection of the cohort; participants in this cohort are then measured or evaluated to determine their exposure status. However, some cohort studies may recruit or select exposed participants in a different time or place than unexposed participants, especially retrospective cohort studies–which is when data are obtained from the past (retrospectively), but the analysis examines exposures prior to outcomes. For example, one research question could be whether diabetic men with clinical depression are at higher risk for cardiovascular disease than those without clinical depression. So, diabetic men with depression might be selected from a mental health clinic, while diabetic men without depression might be selected from an internal medicine or endocrinology clinic. This study recruits groups from different clinic populations, so this example would get a "no."

However, the women nurses described in the question above were selected based on the same inclusion/exclusion criteria, so that example would get a "yes."

Question 5. Sample size justification

Did the authors present their reasons for selecting or recruiting the number of people included or analyzed? Do they note or discuss the statistical power of the study? This question is about whether or not the study had enough participants to detect an association if one truly existed.

A paragraph in the methods section of the article may explain the sample size needed to detect a hypothesized difference in outcomes. You may also find a discussion of power in the discussion section (such as the study had 85 percent power to detect a 20 percent increase in the rate of an outcome of interest, with a 2-sided alpha of 0.05). Sometimes estimates of variance and/or estimates of effect size are given, instead of sample size calculations. In any of these cases, the answer would be "yes."

However, observational cohort studies often do not report anything about power or sample sizes because the analyses are exploratory in nature. In this case, the answer would be "no." This is not a "fatal flaw." It just may indicate that attention was not paid to whether the study was sufficiently sized to answer a prespecified question–i.e., it may have been an exploratory, hypothesis-generating study.

Question 6. Exposure assessed prior to outcome measurement

This question is important because, in order to determine whether an exposure causes an outcome, the exposure must come before the outcome.

For some prospective cohort studies, the investigator enrolls the cohort and then determines the exposure status of various members of the cohort (large epidemiological studies like Framingham used this approach). However, for other cohort studies, the cohort is selected based on its exposure status, as in the example above of depressed diabetic men (the exposure being depression). Other examples include a cohort identified by its exposure to fluoridated drinking water and then compared to a cohort living in an area without fluoridated water, or a cohort of military personnel exposed to combat in the Gulf War compared to a cohort of military personnel not deployed in a combat zone.

With either of these types of cohort studies, the cohort is followed forward in time (i.e., prospectively) to assess the outcomes that occurred in the exposed members compared to nonexposed members of the cohort. Therefore, you begin the study in the present by looking at groups that were exposed (or not) to some biological or behavioral factor, intervention, etc., and then you follow them forward in time to examine outcomes. If a cohort study is conducted properly, the answer to this question should be "yes," since the exposure status of members of the cohort was determined at the beginning of the study before the outcomes occurred.

For retrospective cohort studies, the same principal applies. The difference is that, rather than identifying a cohort in the present and following them forward in time, the investigators go back in time (i.e., retrospectively) and select a cohort based on their exposure status in the past and then follow them forward to assess the outcomes that occurred in the exposed and nonexposed cohort members. Because in retrospective cohort studies the exposure and outcomes may have already occurred (it depends on how long they follow the cohort), it is important to make sure that the exposure preceded the outcome.

Sometimes cross-sectional studies are conducted (or cross-sectional analyses of cohort-study data), where the exposures and outcomes are measured during the same timeframe. As a result, cross-sectional analyses provide weaker evidence than regular cohort studies regarding a potential causal relationship between exposures and outcomes. For cross-sectional analyses, the answer to Question 6 should be "no."

Question 7. Sufficient timeframe to see an effect

Did the study allow enough time for a sufficient number of outcomes to occur or be observed, or enough time for an exposure to have a biological effect on an outcome? In the examples given above, if clinical depression has a biological effect on increasing risk for CVD, such an effect may take years. In the other example, if higher dietary sodium increases BP, a short timeframe may be sufficient to assess its association with BP, but a longer timeframe would be needed to examine its association with heart attacks.

The issue of timeframe is important to enable meaningful analysis of the relationships between exposures and outcomes to be conducted. This often requires at least several years, especially when looking at health outcomes, but it depends on the research question and outcomes being examined.

Cross-sectional analyses allow no time to see an effect, since the exposures and outcomes are assessed at the same time, so those would get a "no" response.

Question 8. Different levels of the exposure of interest

If the exposure can be defined as a range (examples: drug dosage, amount of physical activity, amount of sodium consumed), were multiple categories of that exposure assessed? (for example, for drugs: not on the medication, on a low dose, medium dose, high dose; for dietary sodium, higher than average U.S. consumption, lower than recommended consumption, between the two). Sometimes discrete categories of exposure are not used, but instead exposures are measured as continuous variables (for example, mg/day of dietary sodium or BP values).

In any case, studying different levels of exposure (where possible) enables investigators to assess trends or dose-response relationships between exposures and outcomes–e.g., the higher the exposure, the greater the rate of the health outcome. The presence of trends or dose-response relationships lends credibility to the hypothesis of causality between exposure and outcome.

For some exposures, however, this question may not be applicable (e.g., the exposure may be a dichotomous variable like living in a rural setting versus an urban setting, or vaccinated/not vaccinated with a one-time vaccine). If there are only two possible exposures (yes/no), then this question should be given an "NA," and it should not count negatively towards the quality rating.

Question 9. Exposure measures and assessment

Were the exposure measures defined in detail? Were the tools or methods used to measure exposure accurate and reliable–for example, have they been validated or are they objective? This issue is important as it influences confidence in the reported exposures. When exposures are measured with less accuracy or validity, it is harder to see an association between exposure and outcome even if one exists. Also as important is whether the exposures were assessed in the same manner within groups and between groups; if not, bias may result.

For example, retrospective self-report of dietary salt intake is not as valid and reliable as prospectively using a standardized dietary log plus testing participants' urine for sodium content. Another example is measurement of BP, where there may be quite a difference between usual care, where clinicians measure BP however it is done in their practice setting (which can vary considerably), and use of trained BP assessors using standardized equipment (e.g., the same BP device which has been tested and calibrated) and a standardized protocol (e.g., patient is seated for 5 minutes with feet flat on the floor, BP is taken twice in each arm, and all four measurements are averaged). In each of these cases, the former would get a "no" and the latter a "yes."

Here is a final example that illustrates the point about why it is important to assess exposures consistently across all groups: If people with higher BP (exposed cohort) are seen by their providers more frequently than those without elevated BP (nonexposed group), it also increases the chances of detecting and documenting changes in health outcomes, including CVD-related events. Therefore, it may lead to the conclusion that higher BP leads to more CVD events. This may be true, but it could also be due to the fact that the subjects with higher BP were seen more often; thus, more CVD-related events were detected and documented simply because they had more encounters with the health care system. Thus, it could bias the results and lead to an erroneous conclusion.

Question 10. Repeated exposure assessment

Was the exposure for each person measured more than once during the course of the study period? Multiple measurements with the same result increase our confidence that the exposure status was correctly classified. Also, multiple measurements enable investigators to look at changes in exposure over time, for example, people who ate high dietary sodium throughout the followup period, compared to those who started out high then reduced their intake, compared to those who ate low sodium throughout. Once again, this may not be applicable in all cases. In many older studies, exposure was measured only at baseline. However, multiple exposure measurements do result in a stronger study design.

Question 11. Outcome measures

Were the outcomes defined in detail? Were the tools or methods for measuring outcomes accurate and reliable–for example, have they been validated or are they objective? This issue is important because it influences confidence in the validity of study results. Also important is whether the outcomes were assessed in the same manner within groups and between groups.

An example of an outcome measure that is objective, accurate, and reliable is death–the outcome measured with more accuracy than any other. But even with a measure as objective as death, there can be differences in the accuracy and reliability of how death was assessed by the investigators. Did they base it on an autopsy report, death certificate, death registry, or report from a family member? Another example is a study of whether dietary fat intake is related to blood cholesterol level (cholesterol level being the outcome), and the cholesterol level is measured from fasting blood samples that are all sent to the same laboratory. These examples would get a "yes." An example of a "no" would be self-report by subjects that they had a heart attack, or self-report of how much they weigh (if body weight is the outcome of interest).

Similar to the example in Question 9, results may be biased if one group (e.g., people with high BP) is seen more frequently than another group (people with normal BP) because more frequent encounters with the health care system increases the chances of outcomes being detected and documented.

Question 12. Blinding of outcome assessors

Blinding means that outcome assessors did not know whether the participant was exposed or unexposed. It is also sometimes called "masking." The objective is to look for evidence in the article that the person(s) assessing the outcome(s) for the study (for example, examining medical records to determine the outcomes that occurred in the exposed and comparison groups) is masked to the exposure status of the participant. Sometimes the person measuring the exposure is the same person conducting the outcome assessment. In this case, the outcome assessor would most likely not be blinded to exposure status because they also took measurements of exposures. If so, make a note of that in the comments section.

As you assess this criterion, think about whether it is likely that the person(s) doing the outcome assessment would know (or be able to figure out) the exposure status of the study participants. If the answer is no, then blinding is adequate. An example of adequate blinding of the outcome assessors is to create a separate committee, whose members were not involved in the care of the patient and had no information about the study participants' exposure status. The committee would then be provided with copies of participants' medical records, which had been stripped of any potential exposure information or personally identifiable information. The committee would then review the records for prespecified outcomes according to the study protocol. If blinding was not possible, which is sometimes the case, mark "NA" and explain the potential for bias.

Question 13. Followup rate

Higher overall followup rates are always better than lower followup rates, even though higher rates are expected in shorter studies, whereas lower overall followup rates are often seen in studies of longer duration. Usually, an acceptable overall followup rate is considered 80 percent or more of participants whose exposures were measured at baseline. However, this is just a general guideline. For example, a 6-month cohort study examining the relationship between dietary sodium intake and BP level may have over 90 percent followup, but a 20-year cohort study examining effects of sodium intake on stroke may have only a 65 percent followup rate.

Question 14. Statistical analyses

Were key potential confounding variables measured and adjusted for, such as by statistical adjustment for baseline differences? Logistic regression or other regression methods are often used to account for the influence of variables not of interest.

This is a key issue in cohort studies, because statistical analyses need to control for potential confounders, in contrast to an RCT, where the randomization process controls for potential confounders. All key factors that may be associated both with the exposure of interest and the outcome–that are not of interest to the research question–should be controlled for in the analyses.

For example, in a study of the relationship between cardiorespiratory fitness and CVD events (heart attacks and strokes), the study should control for age, BP, blood cholesterol, and body weight, because all of these factors are associated both with low fitness and with CVD events. Well-done cohort studies control for multiple potential confounders.

Some general guidance for determining the overall quality rating of observational cohort and cross-sectional studies

The questions on the form are designed to help you focus on the key concepts for evaluating the internal validity of a study. They are not intended to create a list that you simply tally up to arrive at a summary judgment of quality.

Internal validity for cohort studies is the extent to which the results reported in the study can truly be attributed to the exposure being evaluated and not to flaws in the design or conduct of the study–in other words, the ability of the study to draw associative conclusions about the effects of the exposures being studied on outcomes. Any such flaws can increase the risk of bias.

Critical appraisal involves considering the risk of potential for selection bias, information bias, measurement bias, or confounding (the mixture of exposures that one cannot tease out from each other). Examples of confounding include co-interventions, differences at baseline in patient characteristics, and other issues throughout the questions above. High risk of bias translates to a rating of poor quality. Low risk of bias translates to a rating of good quality. (Thus, the greater the risk of bias, the lower the quality rating of the study.)

In addition, the more attention in the study design to issues that can help determine whether there is a causal relationship between the exposure and outcome, the higher quality the study. These include exposures occurring prior to outcomes, evaluation of a dose-response gradient, accuracy of measurement of both exposure and outcome, sufficient timeframe to see an effect, and appropriate control for confounding–all concepts reflected in the tool.

Generally, when you evaluate a study, you will not see a "fatal flaw," but you will find some risk of bias. By focusing on the concepts underlying the questions in the quality assessment tool, you should ask yourself about the potential for bias in the study you are critically appraising. For any box where you check "no" you should ask, "What is the potential risk of bias resulting from this flaw in study design or execution?" That is, does this factor cause you to doubt the results that are reported in the study or doubt the ability of the study to accurately assess an association between exposure and outcome?

The best approach is to think about the questions in the tool and how each one tells you something about the potential for bias in a study. The more you familiarize yourself with the key concepts, the more comfortable you will be with critical appraisal. Examples of studies rated good, fair, and poor are useful, but each study must be assessed on its own based on the details that are reported and consideration of the concepts for minimizing bias.

Appendix 8: Mixed Methods Appraisal Tool (MMAT) guidance and scoring

MIXED METHODS APPRAISAL TOOL (MMAT)
VERSION 2018
User guide

Prepared by
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Department of Family Medicine
Département de médecine de famille
Academic excellence and innovation in care, teaching and research
Innovation et excellence académique dans les soins, l'enseignement et la recherche

Last update: August 1st, 2018

What is the MMAT?
The MMAT is a critical appraisal tool that is designed for the appraisal stage of systematic mixed studies reviews, i.e., reviews that include qualitative, quantitative and mixed methods studies. It permits to appraise the methodological quality of five categories to studies: qualitative research, randomized controlled trials, non-randomized studies, quantitative descriptive studies, and mixed methods studies.

How was the MMAT developed?
The MMAT was developed in 2006 (Pluye et al., 2009a) and was revised in 2011 (Pace et al., 2012). The present version 2018 was developed on the basis of findings from a literature review of critical appraisal tools, interviews with MMAT users, and an e-Delphi study with international experts (Hong, 2018). The MMAT developers are continuously seeking for improvement and testing of this tool. Users' feedback is always appreciated.

What the MMAT can be used for?
The MMAT can be used to appraise the quality of empirical studies, i.e., primary research based on experiment, observation or simulation (Abbott, 1998; Porta et al., 2014). It cannot be used for non-empirical papers such as review and theoretical papers. Also, the MMAT allows the appraisal of most common types of study methodologies and designs. However, some specific designs such as economic and diagnostic accuracy studies cannot be assessed with the MMAT. Other critical appraisal tools might be relevant for these designs.

What are the requirements?
Because critical appraisal is about judgment making, it is advised to have at least two reviewers independently involved in the appraisal process. Also, using the MMAT requires experience or training in these domains. For instance, MMAT users may be helped by a colleague with specific expertise when needed.

How to use the MMAT?
This document comprises two parts: checklist (Part I) and explanation of the criteria (Part II).

1. Respond to the two screening questions. Responding 'No' or 'Can't tell' to one or both questions might indicate that the paper is not an empirical study, and thus cannot be appraised using the MMAT. MMAT users might decide not to use these questions, especially if the selection criteria of their review are limited to empirical studies.
2. For each included study, choose the appropriate category of studies to appraise. Look at the description of the methods used in the included studies. If needed, use the algorithm at the end of this document.
3. Rate the criteria of the chosen category. For example, if the paper is a qualitative study, only rate the five criteria in the qualitative category. The 'Can't tell' response category means that the paper do not report appropriate information to answer 'Yes' or 'No', or that report unclear information related to the criterion. Rating 'Can't tell' could lead to look for companion papers, or contact authors to ask more information or clarification when needed. In Part II of this document, indicators are added for some criteria. The list is not exhaustive and not all indicators are necessary. You should agree among your team which ones are important to consider for your field and apply them uniformly across all included studies from the same category.

How to score?
It is discouraged to calculate an overall score from the ratings of each criterion. Instead, it is advised to provide a more detailed presentation of the ratings of each criterion to better inform the quality of the included studies. This may lead to perform a sensitivity analysis (i.e., to consider the quality of studies by contrasting their results). Excluding studies with low methodological quality is usually discouraged.

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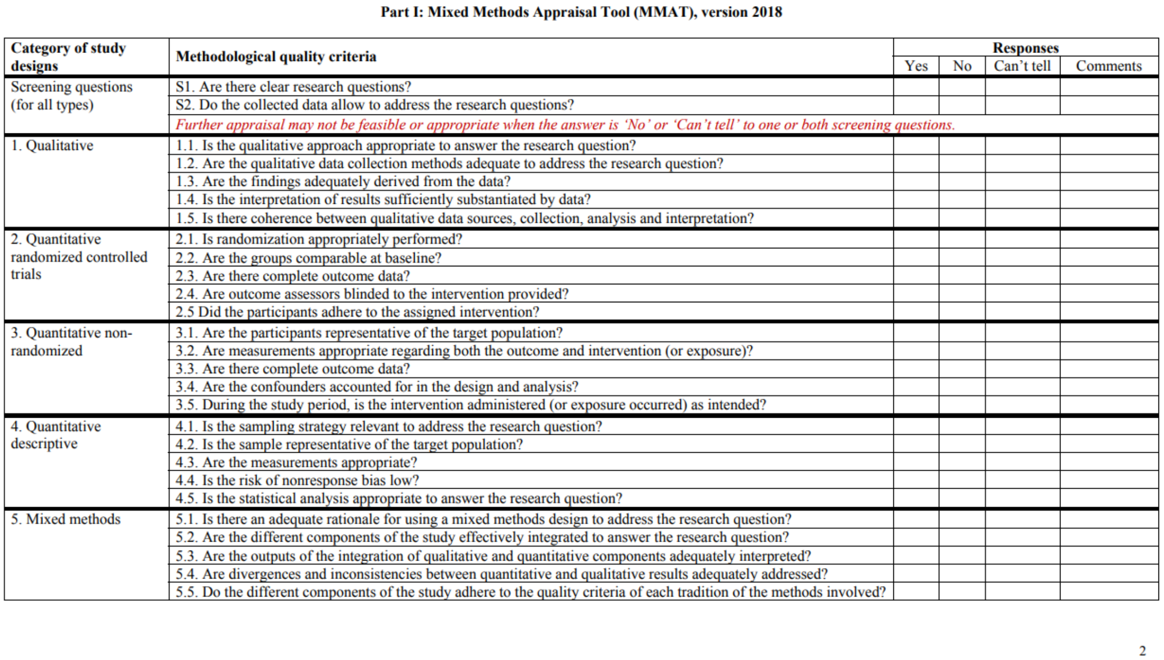
For dissemination, application, and feedback: Please contact mixed.methods.appraisal.tool@gmail.com
For more information: http://mixedmethodsappraisaltoolpublic.pbworks.com/



Part I: Mixed Methods Appraisal Tool (MMAT), version 2018

Category of study designs	**Methodological quality criteria**	**Responses**			
		Yes	No	Can't tell	Comments
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 randomized 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-randomized	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 nonresponse 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 answer 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?				

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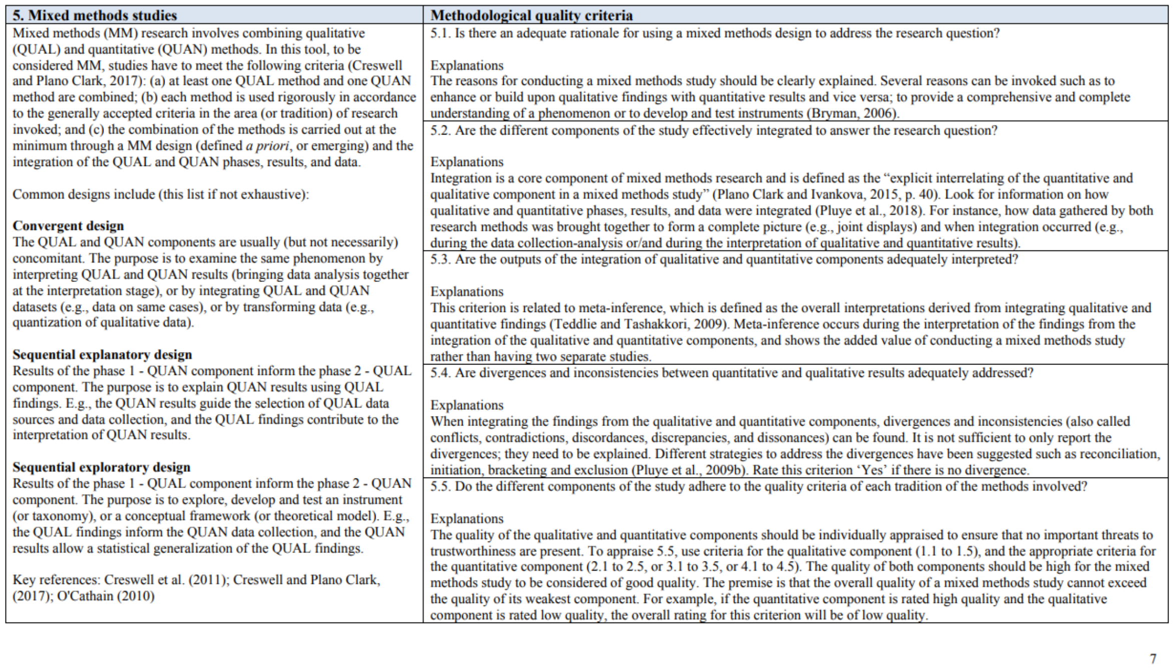
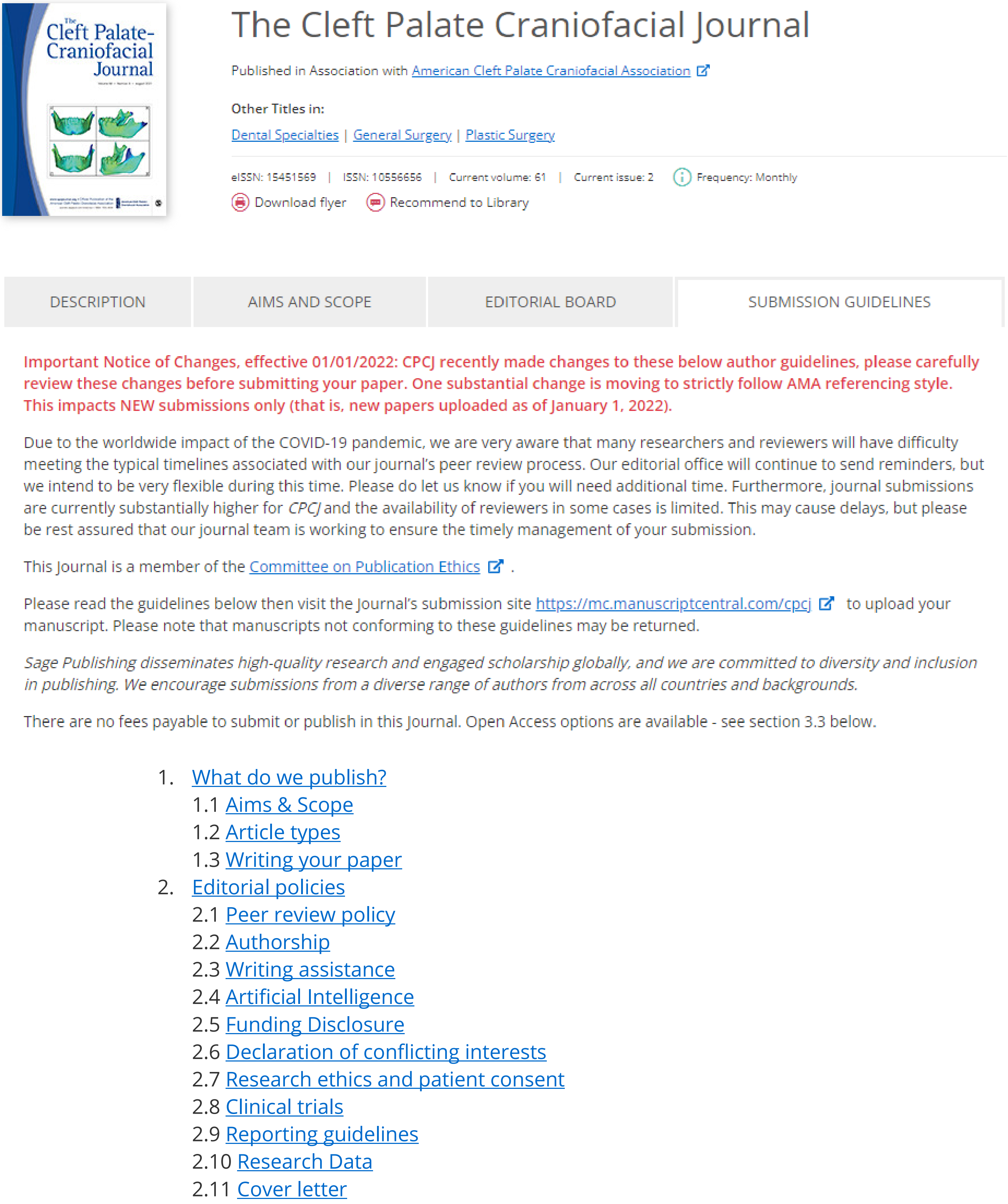


5. Mixed methods studies	**Methodological quality criteria**
Mixed methods (MM) research involves combining qualitative (QUAL) and quantitative (QUAN) methods. In this tool, to be considered MM, studies have to meet the following criteria (Creswell and Plano Clark, 2017): (a) at least one QUAL method and one QUAN method are combined; (b) each method is used rigorously in accordance to the generally accepted criteria in the area (or tradition) of research invoked; and (c) the combination of the methods is carried out at the minimum through a MM design (defined *a priori*, or emerging) and the integration of the QUAL and QUAN phases, results, and data. Common designs include (this list if not exhaustive): **Convergent design** The QUAL and QUAN components are usually (but not necessarily) concomitant. The purpose is to examine the same phenomenon by interpreting QUAL and QUAN results (bringing data analysis together at the interpretation stage), or by integrating QUAL and QUAN datasets (e.g., data on same cases), or by transforming data (e.g., quantization of qualitative data). **Sequential explanatory design** Results of the phase 1 - QUAN component inform the phase 2 - QUAL component. The purpose is to explain QUAN results using QUAL findings. E.g., the QUAN results guide the selection of QUAL data sources and data collection, and the QUAL findings contribute to the interpretation of QUAN results. **Sequential exploratory design** Results of the phase 1 - QUAL component inform the phase 2 - QUAN component. The purpose is to explore, develop and test an instrument (or taxonomy), or a conceptual framework (or theoretical model). E.g., the QUAL findings inform the QUAN data collection, and the QUAN results allow a statistical generalization of the QUAL findings. Key references: Creswell et al. (2011); Creswell and Plano Clark, (2017); O'Cathain (2010)	5.1. Is there an adequate rationale for using a mixed methods design to address the research question? Explanations The reasons for conducting a mixed methods study should be clearly explained. Several reasons can be invoked such as to enhance or build upon qualitative findings with quantitative results and vice versa; to provide a comprehensive and complete understanding of a phenomenon or to develop and test instruments (Bryman, 2006).
	5.2. Are the different components of the study effectively integrated to answer the research question? Explanations Integration is a core component of mixed methods research and is defined as the "explicit interrelating of the quantitative and qualitative component in a mixed methods study" (Plano Clark and Ivankova, 2015, p. 40). Look for information on how qualitative and quantitative phases, results, and data were integrated (Pluye et al., 2018). For instance, how data gathered by both research methods was brought together to form a complete picture (e.g., joint displays) and when integration occurred (e.g., during the data collection-analysis or/and during the interpretation of qualitative and quantitative results).
	5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted? Explanations This criterion is related to meta-inference, which is defined as the overall interpretations derived from integrating qualitative and quantitative findings (Teddlie and Tashakkori, 2009). Meta-inference occurs during the interpretation of the findings from the integration of the qualitative and quantitative components, and shows the added value of conducting a mixed methods study rather than having two separate studies.
	5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed? Explanations When integrating the findings from the qualitative and quantitative components, divergences and inconsistencies (also called conflicts, contradictions, discordances, discrepancies, and dissonances) can be found. It is not sufficient to only report the divergences; they need to be explained. Different strategies to address the divergences have been suggested such as reconciliation, initiation, bracketing and exclusion (Pluye et al., 2009b). Rate this criterion 'Yes' if there is no divergence.
	5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved? Explanations The quality of the qualitative and quantitative components should be individually appraised to ensure that no important threats to trustworthiness are present. To appraise 5.5, use criteria for the qualitative component (1.1 to 1.5), and the appropriate criteria for the quantitative component (2.1 to 2.5, or 3.1 to 3.5, or 4.1 to 4.5). The quality of both components should be high for the mixed methods study to be considered of good quality. The premise is that the overall quality of a mixed methods study cannot exceed the quality of its weakest component. For example, if the quantitative component is rated high quality and the qualitative component is rated low quality, the overall rating for this criterion will be of low quality.

7

Appendix 9. The Cleft Palate-Craniofacial Journal submission guidelines



The Cleft Palate Craniofacial Journal

Published in Association with American Cleft Palate Craniofacial Association

Other Titles in:

Dental Specialties | General Surgery | Plastic Surgery

eISSN: 15451569 | ISSN: 10556656 | Current volume: 61 | Current issue: 2 | Frequency: Monthly

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Important Notice of Changes, effective 01/01/2022: CPCJ recently made changes to these below author guidelines, please carefully review these changes before submitting your paper. One substantial change is moving to strictly follow AMA referencing style. This impacts NEW submissions only (that is, new papers uploaded as of January 1, 2022).

Due to the worldwide impact of the COVID-19 pandemic, we are very aware that many researchers and reviewers will have difficulty meeting the typical timelines associated with our journal's peer review process. Our editorial office will continue to send reminders, but we intend to be very flexible during this time. Please do let us know if you will need additional time. Furthermore, journal submissions are currently substantially higher for *CPCJ* and the availability of reviewers in some cases is limited. This may cause delays, but please be rest assured that our journal team is working to ensure the timely management of your submission.

This Journal is a member of the Committee on Publication Ethics .

Please read the guidelines below then visit the Journal's submission site https://mc.manuscriptcentral.com/cpcj to upload your manuscript. Please note that manuscripts not conforming to these guidelines may be returned.

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There are no fees payable to submit or publish in this Journal. Open Access options are available - see section 3.3 below.

1. What do we publish?
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    2.10 Research Data
    2.11 Cover letter

Research

PRISMA checklist
CONSORT checklist

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

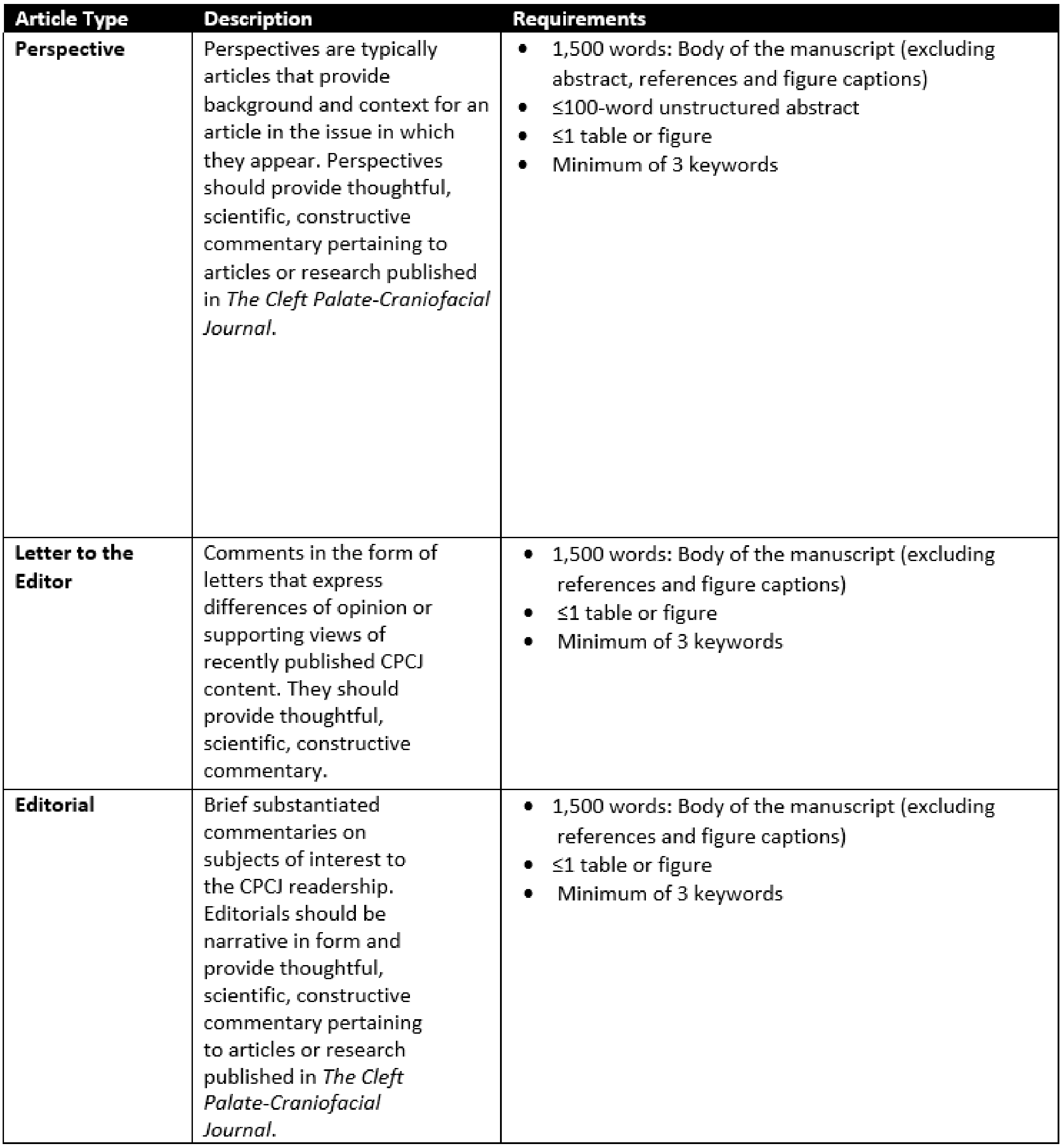
What I (We) Do	Introduce new solutions to clinical problems. Novelty and quality of illustrations and videos (when appropriate) are key ingredients. If no patient identifiable data are included, no IRB form is necessary.	• 1,000 words: Body of the manuscript (excluding abstract, references and figure captions) • 50–75-word structured abstract with the following format: background (what is the issue/problem), solution, what I/we did that is new • 3-5 keywords • ≤3 tables and figures, combined • ≤5 references
Case/Clinical Reports	Case reports presenting new clinical information.	• 4,000 words: Body of the manuscript (excluding abstract, references and figure captions) • ≤100-word unstructured abstract, describing the objective, essential features and uniqueness of the case being presented, and conclusions • Minimum of 3 keywords • ≤6 tables or figures, combined

Ethics/Health Policy	Ethical and Legal Reports are original articles which examine issues of ethics or the law arising in cleft and craniofacial care and research. Health Policy Reports are original articles which examine social, political, and economic issues arising in cleft and craniofacial care or research.	• 3,000 words: Body of the manuscript (excluding abstract, references and figure captions) • ≤100-word unstructured abstract • Minimum of 3 keywords • ≤3 figures and tables, combined
Brief Communications	Preliminary or limited results of original research pertaining to prevalence, causes, mechanisms, diagnosis, course, treatment, and prevention.	• 3,000 words: Body of the manuscript (excluding abstract, references and figure captions) • 150-word structured abstract using the below headings: • Objective • Design • Setting • Patients, Participants • Interventions • Main Outcome Measure(s) • Results • Conclusions • Minimum of 3 keywords • For non-data brief communications, ≤100-word unstructured abstract • ≤3 figures and tables, combined

Clinical Review and Education

Article Type	Description	Requirements
Ideas and Innovations	Short communications related to novel ideas, techniques, methods of assessment, etc.	• 3,000 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 • ≤3 figures and tables, combined

Opinion



Article Type	Description	Requirements
Perspective	Perspectives are typically articles that provide background and context for an article in the issue in which they appear. Perspectives should provide thoughtful, scientific, constructive commentary pertaining to articles or research published in *The Cleft Palate-Craniofacial Journal*.	• 1,500 words: Body of the manuscript (excluding abstract, references and figure captions) • ≤100-word unstructured abstract • ≤1 table or figure • Minimum of 3 keywords
Letter to the Editor	Comments in the form of letters that express differences of opinion or supporting views of recently published CPCJ content. They should provide thoughtful, scientific, constructive commentary.	• 1,500 words: Body of the manuscript (excluding references and figure captions) • ≤1 table or figure • Minimum of 3 keywords
Editorial	Brief substantiated commentaries on subjects of interest to the CPCJ readership. Editorials should be narrative in form and provide thoughtful, scientific, constructive commentary pertaining to articles or research published in *The Cleft Palate-Craniofacial Journal*.	• 1,500 words: Body of the manuscript (excluding references and figure captions) • ≤1 table or figure • Minimum of 3 keywords

1.3 Writing your paper

The Sage Author Gateway has some general advice and on how to get published, plus links to further resources. Sage Author Services also offers authors a variety of ways to improve and enhance their article including English language editing, plagiarism detection, and video abstract and infographic preparation.

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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.

Two independent peer reviews are typically solicited. At the discretion of the Section Editor, a third review may be requested and/or a review by a biostatistician may also be solicited. The Editor is responsible for all final decisions regarding acceptance or rejection, recommendations for revision, and final editing. Manuscripts will be evaluated according to various criteria, including scientific methodology, level of evidence, novelty, clarity, and conciseness. Accepted articles describing novel findings or methods with high levels of evidence may be advanced in the publication queue at the discretion of the Editor.

All submitted articles are "double-anonymized" to ensure an unbiased review. Reviewers will not have access to author names or affiliations. Authors will not have access to reviewer names or affiliations.

The Editor or members of the Editorial Board may occasionally submit their own manuscripts for possible publication in the journal. In these cases, the peer review process will be managed by alternative members of the Board and the submitting Editor/Board member will have no involvement in the decision-making process.

CPCJ is committed to delivering high quality, fast peer-review for your paper, and as such has partnered with Publons. Publons is a third-party service that seeks to track, verify and give credit for peer review. Reviewers for *CPCJ* can opt in to Publons in order to claim their reviews or have them automatically verified and added to their reviewer profile. Reviewers claiming credit for their review will be associated with the relevant journal, but the article name, reviewer's decision and the content of their review is not published on the site. For more information visit the Publons website.

Reviewers for CPCJ have the option to invite a Reviewer in Training (a graduate student, postdoctoral fellow, early-career research assistant or associate) to serve as a co-reviewer. This program is completely optional. This opportunity is afforded as an educational experience to the Reviewer in Training. The quality of the review is the responsibility of the lead reviewer and not of the Reviewer in Training. The Reviewer in Training will not receive any communications about the manuscript. The use of the contact information for the Reviewer in Training may be used to invite and authorize reviewer roles in the future. If the mentee wishes to be recognized in Publons, the lead reviewer can forward the email that certifies they completed the review to the mentee and then the mentee can send that to Publons. If you worked with a reviewer in training and wish to give them credit, please complete the survey here: https://survey.alchemer.com/s3/7392813/CPCJ-Reviewer-Scoresheet

2.2 Authorship

Papers should only be submitted for consideration once consent is given by all contributing authors. Those submitting papers should carefully check that all those whose work contributed to the paper are acknowledged as contributing authors.

The list of authors should include all those who can legitimately claim authorship. This is all those who:

1. Made a substantial contribution to the concept or design of the work; or acquisition, analysis or interpretation of data,
2. Drafted the article or revised it critically for important intellectual content,
3. Approved the version to be published,
4. Participated sufficiently in the work to take public responsibility for appropriate portions of the content.

The corresponding author must declare his or her contribution to the manuscript by signing the copyright transfer form on behalf of all authors. Authors should meet the conditions of all the points above.

CPCJ follows authorship guidelines as outlined by the International Committee of Medical Journal Editors (ICMJE). Only those involved in writing the paper should be included in the author line. Others should be listed as a footnote or acknowledgment. These authors will be indexed in PubMed as full authors.

The *CPCJ* allows research groups to be recognized in submitted manuscripts. Authors should identify both the group name and the individual authors who accept responsibility for the article (e.g., Smith A, Johnson R, Williams T; The CleftCran Research Group). The named individuals must meet the full criteria and requirements for authorship as described above. Other research group members who do not qualify for authorship may be listed in an Acknowledgement.

Acquisition of funding, collection of data, or general supervision of the research group alone does not constitute authorship, although all contributors who do not meet the criteria for authorship should be listed in the Acknowledgments section. Please refer to the International Committee of Medical Journal Editors (ICMJE) authorship guidelines for more information on authorship.

Authors should determine the order of authorship among themselves and should settle any disagreements before submitting their manuscript. Changes in authorship (ie, order, addition, and deletion of authors) should be discussed and approved by all authors. Any requests for such changes in authorship after initial manuscript submission and before publication should be explained in writing to the editor in a letter or email from all authors.

Please note that AI chatbots, for example ChatGPT, should not be listed as authors. For more information see the policy on Use of ChatGPT and generative AI tools.

2.3 Writing assistance

Individuals who provided writing assistance, e.g., from a specialist communications company, do not qualify as authors and so 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.

It is not necessary to disclose use of language polishing services.

2.4 Artificial Intelligence

Use of Large Language Models and generative AI tools in writing your submission

Sage recognizes the value of large language models (LLMs) (e.g. ChatGPT) and generative AI as productivity tools that can help authors in preparing their article for submission; to generate initial ideas for a structure, for example, or when summarizing, paraphrasing, language polishing etc. However, it is important to note that all language models have limitations and are unable to replicate human creative and critical thinking. Human intervention with these tools is essential to ensure that content presented is accurate and appropriate to the reader. Sage therefore requires authors to be aware of the limitations of language models and to consider these in any use of LLMs in their submissions:

Objectivity: Previously published content that contains racist, sexist or other biases can be present in LLM-generated text, and minority viewpoints may not be represented. Use of LLMs has the potential to perpetuate these biases because the information is decontextualized and harder to detect.
Accuracy: LLMs can 'hallucinate' i.e. generate false content, especially when used outside of their domain or when dealing with complex or ambiguous topics. They can generate content that is linguistically but not scientifically plausible, they can get facts wrong, and they have been shown to generate citations that don't exist. Some LLMs are only trained on content published before a particular date and therefore present an incomplete picture.
Contextual understanding: LLMs cannot apply human understanding to the context of a piece of text, especially when dealing with idiomatic expressions, sarcasm, humor, or metaphorical language. This can lead to errors or misinterpretations in the generated content.
Training data: LLMs require a large amount of high-quality training data to achieve optimal performance. However, in some domains or languages, such data may not be readily available, limiting the usefulness of the model.

Guidance for authors

Authors are required to:

1. **Clearly indicate the use of language models in the manuscript**, including which model was used and for what purpose. Please use the methods or acknowledgements section, as appropriate.
2. **Verify the accuracy, validity, and appropriateness of the content** and any citations generated by language models and correct any errors or inconsistencies.
3. **Provide a list of sources used to generate content** and citations, including those generated by language models. Double-check citations to ensure they are accurate, and are properly referenced.
4. **Be conscious of the potential for plagiarism** where the LLM may have reproduced substantial text from other sources. Check the original sources to be sure you are not plagiarizing someone else's work.

5. **Acknowledge the limitations of language models in the manuscript**, including the potential for bias, errors, and gaps in knowledge.
6. Please note that AI bots such as ChatGPT **should not be listed as an author** on your submission.

We will take appropriate corrective action where we identify published articles with undisclosed use of such tools.

2.5 Funding Disclosure

CPCJ requires all authors to report their funding. Authors will be asked to disclose any sources of funding during submission. Be sure to include all relevant grant numbers and the names of the granting agencies. This information will be used to generate a funding statement that will appear at the end of the manuscript. Funding information should not be included in the acknowledgements or manuscript text because it can compromise anonymity during peer review.

2.6 Declaration of conflicting interests

It is the policy of *CPCJ* to require a declaration of conflicting interests from all authors enabling a statement to be carried within the paginated pages of all published articles. Authors are required to disclose, on the title page included with the submission, 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. This information will be held in confidence by the Editor during the review process but will be included in publication of an accepted manuscript. If no conflict exists, please state that '*The Author(s) declare(s) that there is no conflict of interest*'.

For guidance on conflict of interest statements, please see the ICMJE recommendations here.

2.7 Research ethics and patient consent

Medical research involving human subjects must be conducted according to the World Medical Association Declaration of Helsinki. Compliance with these guidelines should be indicated in the Methods section of the manuscript, along with Institutional Review Board approval if appropriate.

Submitted manuscripts should conform to the ICMJE Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals, and all papers reporting animal and/or human studies must state in the methods section that the relevant Ethics Committee or Institutional Review Board (IRB) provided (or waived) approval. Please ensure that you have provided the full name and institution of the review committee, in addition to the approval number.

While informed consent might not be required for consecutive case series and/or retrospective chart review reports, these are still considered research given that the objective of your report is to generalize the findings. As such, they require Humans Subjects Review Board approval. If IRB approval is not available, the authors must state so in a cover letter accompanying the submission and include a statement in the manuscript that principles outlined in the Declaration of Helsinki were followed.

For research articles, authors are also required to state in the methods section whether participants provided informed consent and whether the consent was written or verbal.

Information on informed consent to report individual cases or case series should be included in the manuscript text. A statement is required regarding whether written informed consent for patient information and images to be published was provided by the patient(s) or a legally authorized representative. The author is responsible for ensuring the anonymity of protection of any individual depicted in a manuscript. A signed permission form must be obtained for any recognizable individual appearing in manuscript figures.

Shading of the eyes is not an acceptable means of rendering an individual unrecognizable. If an author chooses to use his/her own institutional patient permission form, it must include permission to use photographs for all types of publication including but not limited to print, visual, electronic, or broadcast media.

Please also refer to the ICMJE Recommendations for the Protection of Research Participants.

All research involving animals submitted for publication must be approved by an ethics committee with oversight of the facility in which the studies were conducted. The Journal has adopted the ARRIVE guidelines.

2.8 Clinical trials

CPCJ endorses the ICMJE requirement that clinical trials are registered in a WHO-approved public trials registry at or before the time of first patient enrolment. However, consistent with the AllTrials campaign, retrospectively registered trials will be considered if the justification for late registration is acceptable. The trial registry name and URL, and registration number must be included at the end of the abstract.

2.9 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.

Other resources can be found at NLM's Research Reporting Guidelines and Initiatives.

2.10 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.

2.11 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 programme and Open Access agreements, where authors can publish open access either discounted or free of charge depending on the agreement with Sage. Find out if your institution is participating by visiting Open Access Agreements at Sage. For more information on Open Access publishing options at Sage please visit Sage Open Access. For information on funding body compliance, and depositing your article in repositories, please visit Sage's Author Archiving and Re-Use Guidelines and Publishing Policies.

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.

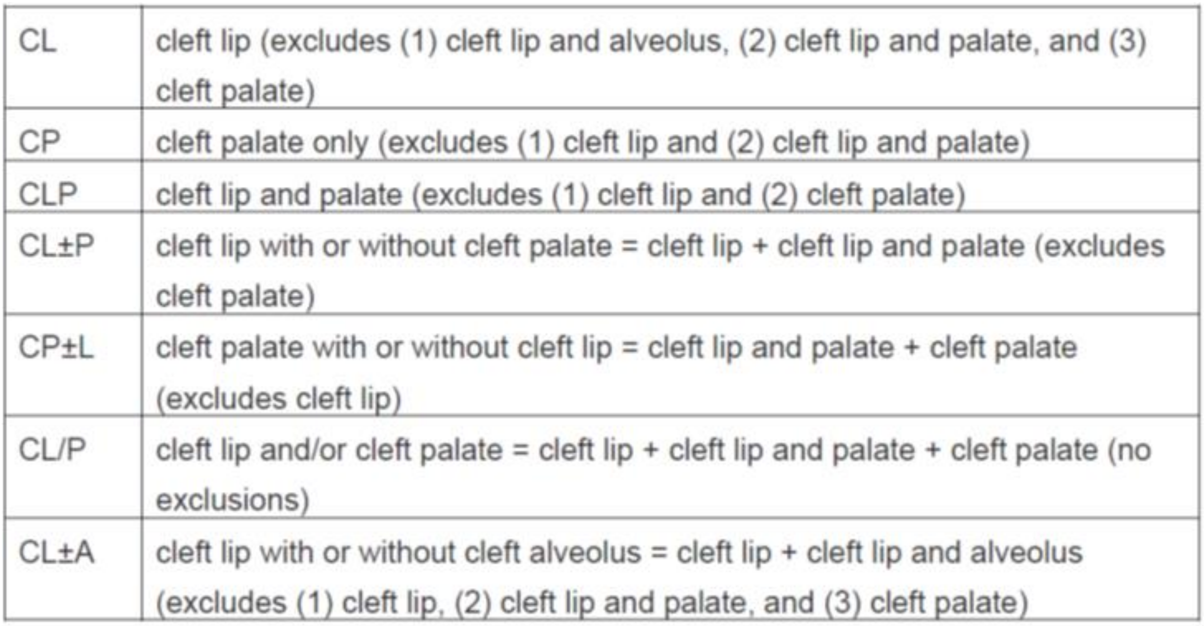
Statistics

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.



CL	cleft lip (excludes (1) cleft lip and alveolus, (2) cleft lip and palate, and (3) cleft palate)
CP	cleft palate only (excludes (1) cleft lip and (2) cleft lip and palate)
CLP	cleft lip and palate (excludes (1) cleft lip and (2) cleft palate)
CL±P	cleft lip with or without cleft palate = cleft lip + cleft lip and palate (excludes cleft palate)
CP±L	cleft palate with or without cleft lip = cleft lip and palate + cleft palate (excludes cleft lip)
CL/P	cleft lip and/or cleft palate = cleft lip + cleft lip and palate + cleft palate (no exclusions)
CL±A	cleft lip with or without cleft alveolus = cleft lip + cleft lip and alveolus (excludes (1) cleft lip, (2) cleft lip and palate, and (3) cleft palate)

Modifying terms that may added to the abbreviations above include:

i (isolated)

I (incomplete)

U (unilateral)

B (bilateral)

SM (submucous)

Phonetic Symbols

Authors who use phonetic symbols are required to use Unicode-compliant fonts in their manuscripts. This will ensure the symbols display properly both during peer review and in the final published article. Examples of acceptable fonts include Charis SIL, Doulos SIL, and Gentium Unicode. Times New Roman is also acceptable, as it includes most IPA symbols and is Unicode compliant.

Citations/References

For citations and references, as of 2022 *CPCJ* uses the 11th Edition AMA Manual of Style. Note that in this style, in-text citations are represented by superscript numerals.

Figure Legends

A list of figure legends must be included on a separate page at the end of the manuscript article file. The legend should explain each figure as concisely as possible. Do not include figure legends in your figure art file. Figure legends are not included in the word count limit.

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.
- **Resolution:** Rasterized based files (i.e., with .tiff or .jpeg extension) require a resolution of at least **300 dpi** (dots per inch). Line art should be supplied with a minimum resolution of **800 dpi**.
- **File size limits: File sizes should be kept below 10MB where possible.**
- **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.

- **Dimension:** Check that the artworks supplied matches or exceeds the dimensions of the journal.
- **Fonts:** The lettering used in the artwork should not vary too much in size and type (usually sans serif font as a default).

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.

If the author does not hold copyright to the video, the author must obtain permission for the video to be published in the journal. This permission must be for unrestricted use in all print, online, and licensed versions of the journal.

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

As part of our commitment to ensuring an ethical, transparent and fair peer review process Sage is a supporting member of ORCID, the Open Researcher and Contributor ID. ORCID provides a unique and persistent digital identifier that distinguishes researchers from every other researcher, even those who share the same name, and, through integration in key research workflows such as manuscript and grant submission, supports automated linkages between researchers and their professional activities, ensuring that their work is recognized.

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.

If you do not already have an ORCID ID please follow this link to create one or visit our ORCID homepage to learn more.

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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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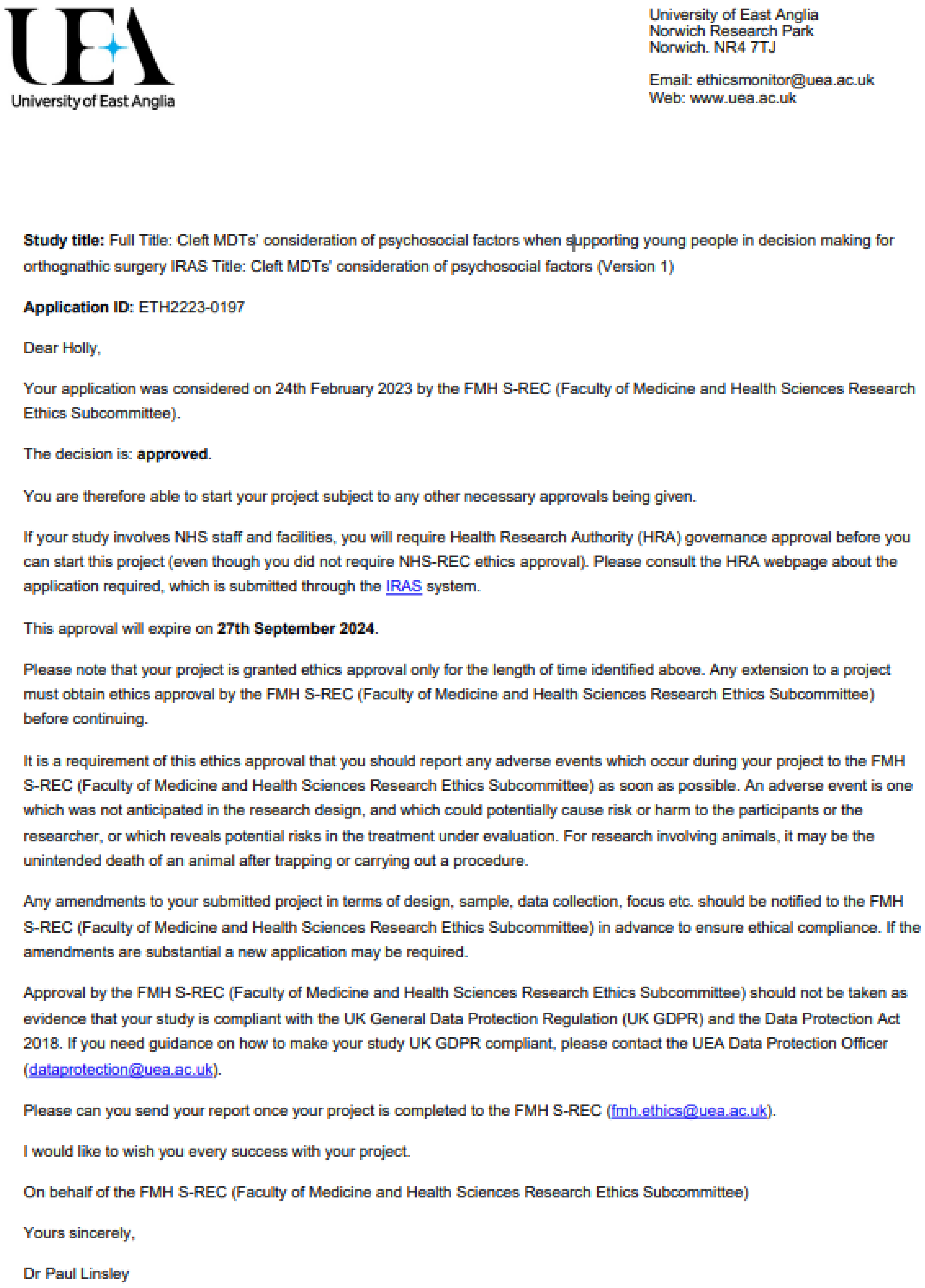
7. Appealing the publication decision

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 10: **UEA FHM S-REC Approval**



University of East Anglia
Norwich Research Park
Norwich. NR4 7TJ

Email: ethicsmonitor@uea.ac.uk
Web: www.uea.ac.uk

Study title: Full Title: Cleft MDTs' consideration of psychosocial factors when supporting young people in decision making for orthognathic surgery IRAS Title: Cleft MDTs' consideration of psychosocial factors (Version 1)

Application ID: ETH2223-0197

Dear Holly,

Your application was considered on 24th February 2023 by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee).

The decision is: **approved**.

You are therefore able to start your project subject to any other necessary approvals being given.

If your study involves NHS staff and facilities, you will require Health Research Authority (HRA) governance approval before you can start this project (even though you did not require NHS-REC ethics approval). Please consult the HRA webpage about the application required, which is submitted through the IRAS system.

This approval will expire on **27th September 2024**.

Please note that your project is granted ethics approval only for the length of time identified above. Any extension to a project must obtain ethics approval by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) before continuing.

It is a requirement of this ethics approval that you should report any adverse events which occur during your project to the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) as soon as possible. An adverse event is one which was not anticipated in the research design, and which could potentially cause risk or harm to the participants or the researcher, or which reveals potential risks in the treatment under evaluation. For research involving animals, it may be the unintended death of an animal after trapping or carrying out a procedure.

Any amendments to your submitted project in terms of design, sample, data collection, focus etc. should be notified to the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) in advance to ensure ethical compliance. If the amendments are substantial a new application may be required.

Approval by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) should not be taken as evidence that your study is compliant with the UK General Data Protection Regulation (UK GDPR) and the Data Protection Act 2018. If you need guidance on how to make your study UK GDPR compliant, please contact the UEA Data Protection Officer (dataprotection@uea.ac.uk).

Please can you send your report once your project is completed to the FMH S-REC (fmh.ethics@uea.ac.uk).

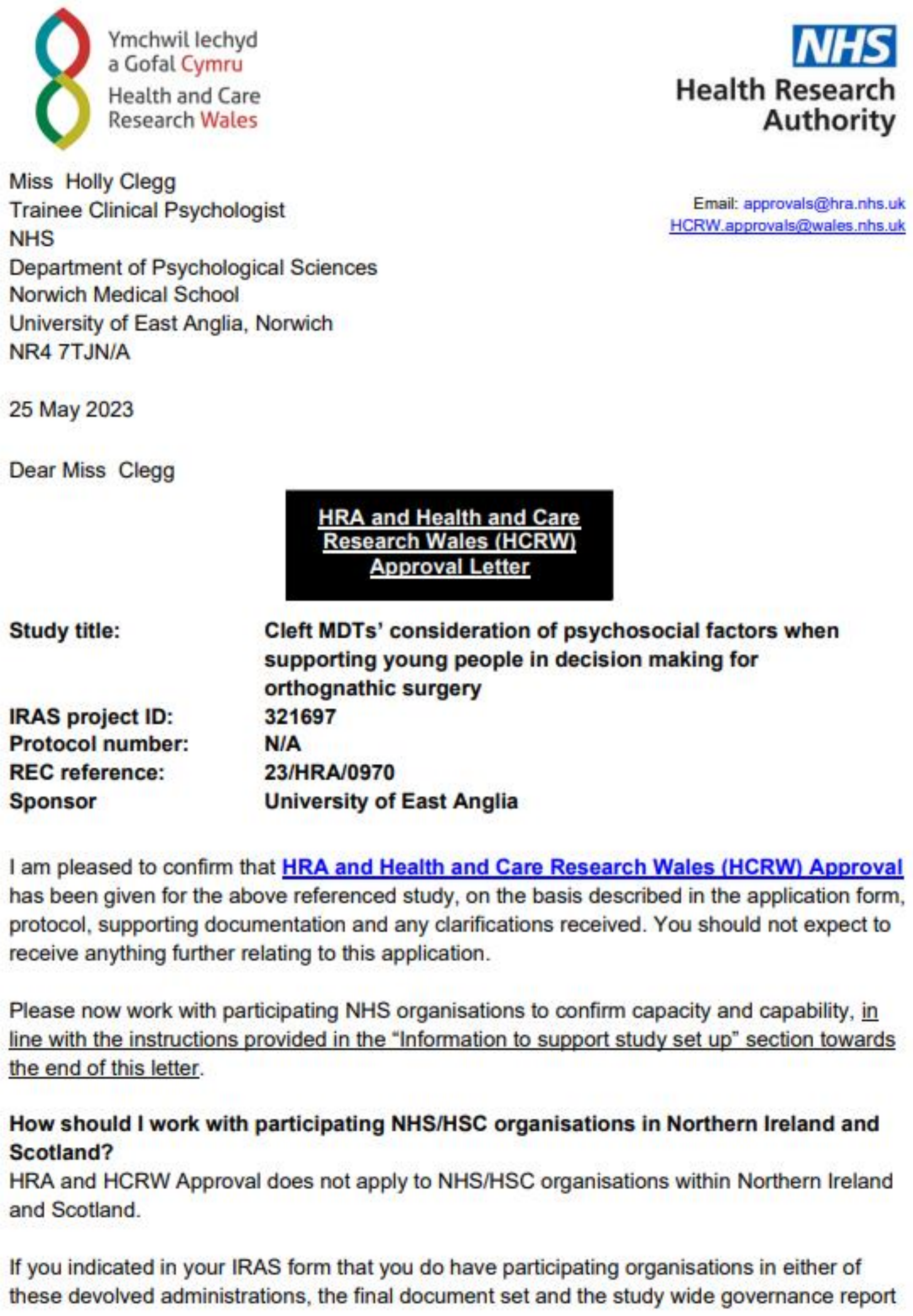
I would like to wish you every success with your project.

On behalf of the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee)

Yours sincerely,

Dr Paul Linsley

Appendix 11: Health Research Authority (HRA) Approval



Ymchwil Iechyd
a Gofal Cymru
Health and Care
Research Wales

NHS
Health Research
Authority

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

Miss Holly Clegg
Trainee Clinical Psychologist
NHS
Department of Psychological Sciences
Norwich Medical School
University of East Anglia, Norwich
NR4 7TJN/A

25 May 2023

Dear Miss Clegg

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Cleft MDTs' consideration of psychosocial factors when supporting young people in decision making for orthognathic surgery**
IRAS project ID:	**321697**
Protocol number:	**N/A**
REC reference:	**23/HRA/0970**
Sponsor	**University of East Anglia**

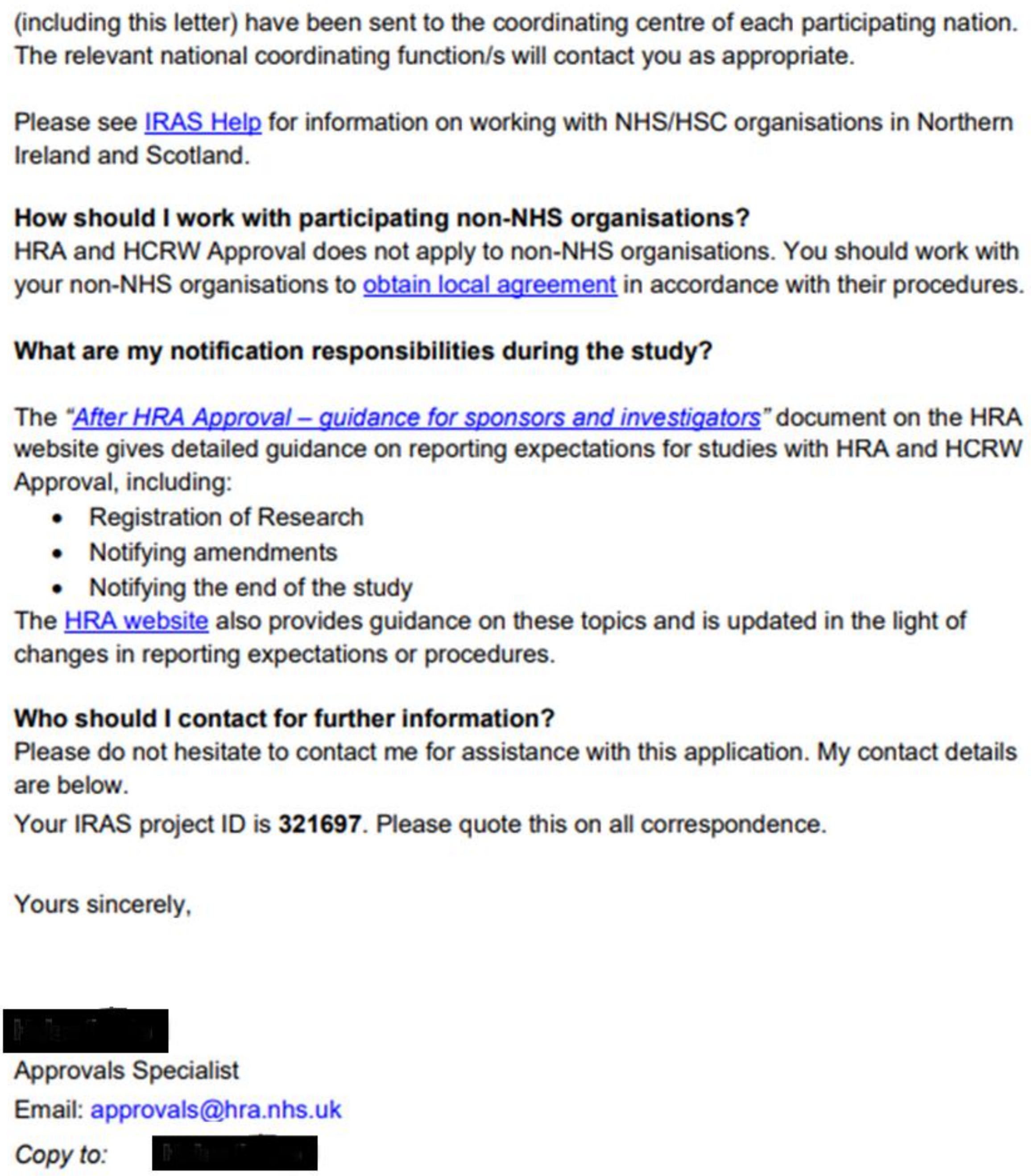
I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report



(including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The *"After HRA Approval – guidance for sponsors and investigators"* document on the HRA website gives detailed guidance on reporting expectations for studies with HRA and HCRW Approval, including:

- Registration of Research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **321697**. Please quote this on all correspondence.

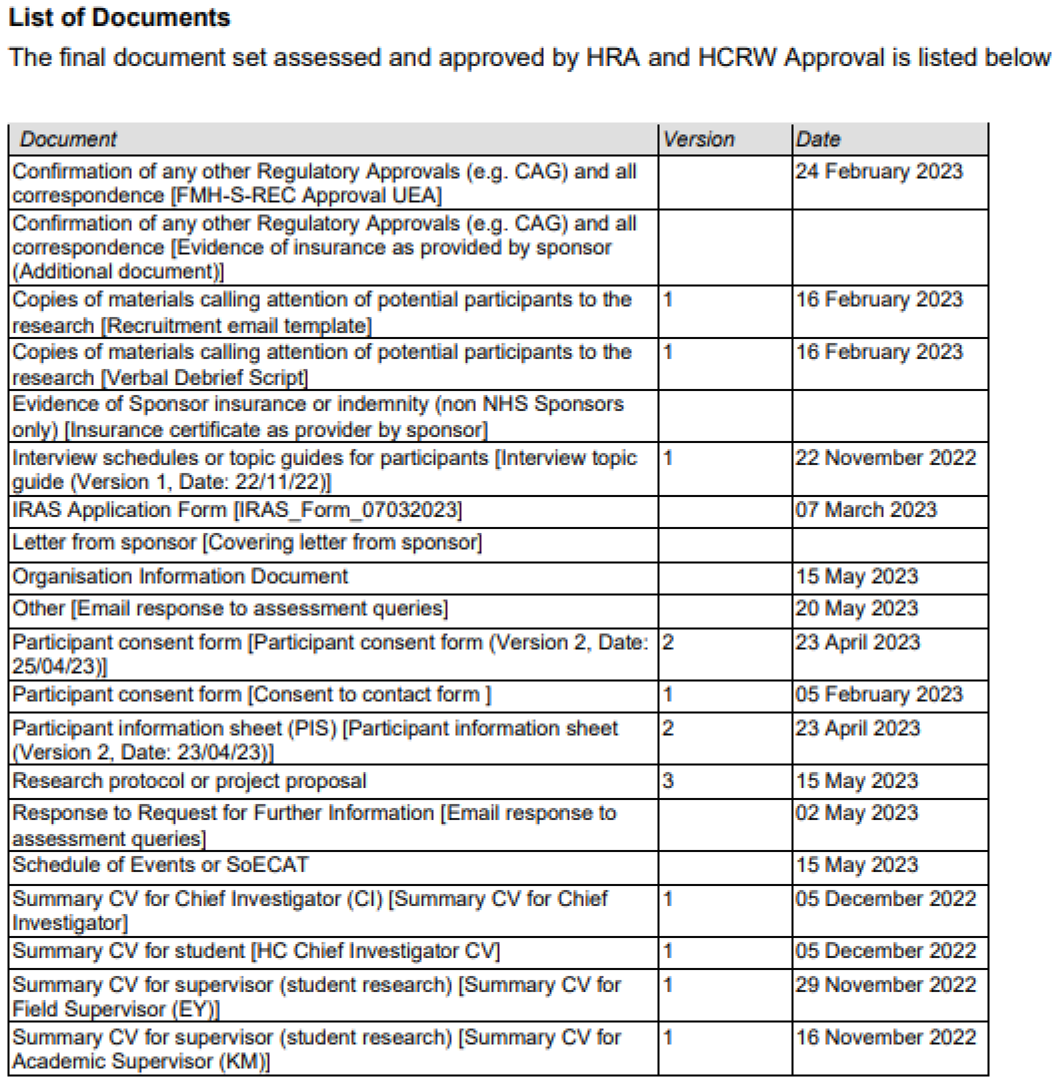
Yours sincerely,

[redacted]

Approvals Specialist

Email: approvals@hra.nhs.uk

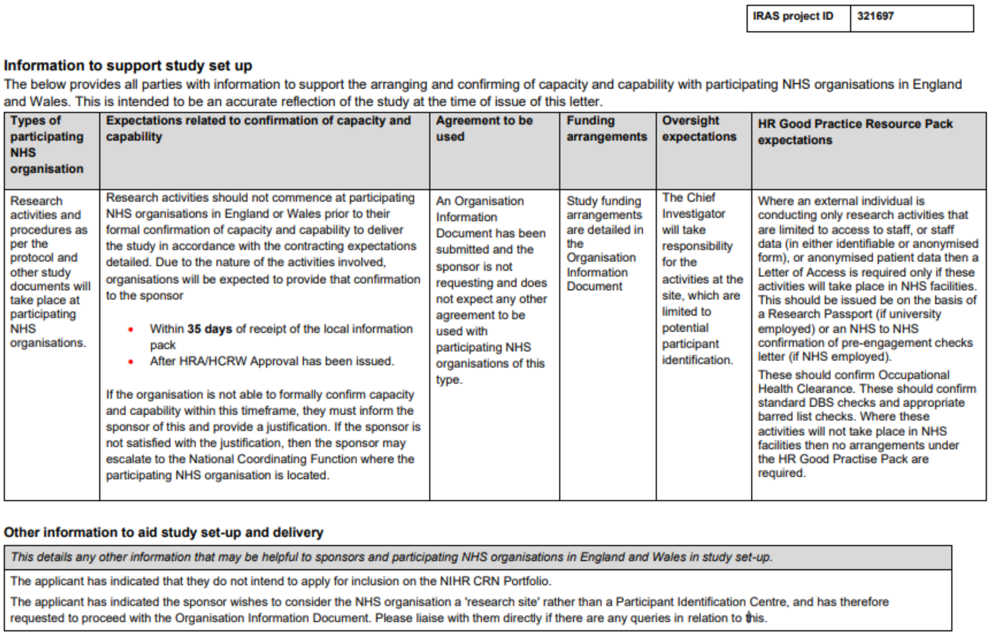
Copy to: [redacted]



List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below

Document	*Version*	*Date*
Confirmation of any other Regulatory Approvals (e.g. CAG) and all correspondence [FMH-S-REC Approval UEA]		24 February 2023
Confirmation of any other Regulatory Approvals (e.g. CAG) and all correspondence [Evidence of insurance as provided by sponsor (Additional document)]		
Copies of materials calling attention of potential participants to the research [Recruitment email template]	1	16 February 2023
Copies of materials calling attention of potential participants to the research [Verbal Debrief Script]	1	16 February 2023
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Insurance certificate as provider by sponsor]		
Interview schedules or topic guides for participants [Interview topic guide (Version 1, Date: 22/11/22)]	1	22 November 2022
IRAS Application Form [IRAS_Form_07032023]		07 March 2023
Letter from sponsor [Covering letter from sponsor]		
Organisation Information Document		15 May 2023
Other [Email response to assessment queries]		20 May 2023
Participant consent form [Participant consent form (Version 2, Date: 25/04/23)]	2	23 April 2023
Participant consent form [Consent to contact form]	1	05 February 2023
Participant information sheet (PIS) [Participant information sheet (Version 2, Date: 23/04/23)]	2	23 April 2023
Research protocol or project proposal	3	15 May 2023
Response to Request for Further Information [Email response to assessment queries]		02 May 2023
Schedule of Events or SoECAT		15 May 2023
Summary CV for Chief Investigator (CI) [Summary CV for Chief Investigator]	1	05 December 2022
Summary CV for student [HC Chief Investigator CV]	1	05 December 2022
Summary CV for supervisor (student research) [Summary CV for Field Supervisor (EY)]	1	29 November 2022
Summary CV for supervisor (student research) [Summary CV for Academic Supervisor (KM)]	1	16 November 2022



IRAS project ID	**321697**

Information to support study set up

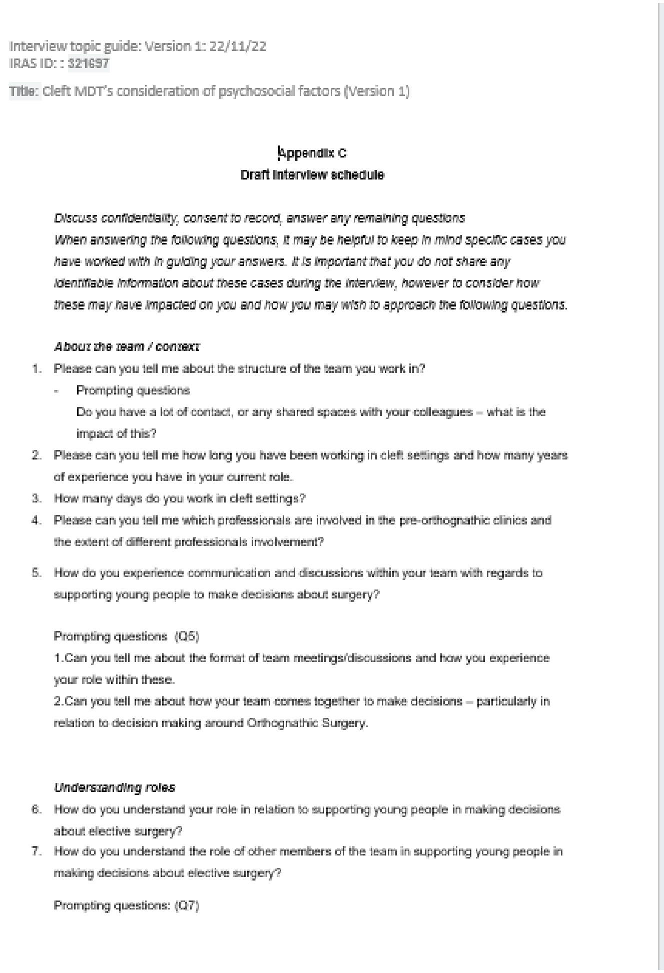
The below provides all parties with information to support the arranging and confirming of capacity and capability with participating NHS organisations in England and Wales. This is intended to be an accurate reflection of the study at the time of issue of this letter.

Types of participating NHS organisation	**Expectations related to confirmation of capacity and capability**	**Agreement to be used**	**Funding arrangements**	**Oversight expectations**	**HR Good Practice Resource Pack expectations**
Research activities and procedures as per the protocol and other study documents will take place at participating NHS organisations.	Research activities should not commence at participating NHS organisations in England or Wales prior to their formal confirmation of capacity and capability to deliver the study in accordance with the contracting expectations detailed. Due to the nature of the activities involved, organisations will be expected to provide that confirmation to the sponsor • Within **35 days** of receipt of the local information pack • After HRA/HCRW Approval has been issued. If the organisation is not able to formally confirm capacity and capability within this timeframe, they must inform the sponsor of this and provide a justification. If the sponsor is not satisfied with the justification, then the sponsor may escalate to the National Coordinating Function where the participating NHS organisation is located.	An Organisation Information Document has been submitted and the sponsor is not requesting and does not expect any other agreement to be used with participating NHS organisations of this type.	Study funding arrangements are detailed in the Organisation Information Document	The Chief Investigator will take responsibility for the activities at the site, which are limited to potential participant identification.	Where an external individual is conducting only research activities that are limited to access to staff, or staff data (in either identifiable or anonymised form), or anonymised patient data then a Letter of Access is required only if these activities will take place in NHS facilities. This should be issued be on the basis of a Research Passport (if university employed) or an NHS to NHS confirmation of pre-engagement checks letter (if NHS employed). These should confirm Occupational Health Clearance. These should confirm standard DBS checks and appropriate barred list checks. Where these activities will not take place in NHS facilities then no arrangements under the HR Good Practise Pack are required.

Other information to aid study set-up and delivery

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.
The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio. The applicant has indicated the sponsor wishes to consider the NHS organisation a 'research site' rather than a Participant Identification Centre, and has therefore requested to proceed with the Organisation Information Document. Please liaise with them directly if there are any queries in relation to this.

Appendix 12: Interview topic guide



Interview topic guide: Version 1: 22/11/22
IRAS ID: : 321697

Title: Cleft MDT's consideration of psychosocial factors (Version 1)

Appendix C
Draft Interview schedule

Discuss confidentiality, consent to record, answer any remaining questions
When answering the following questions, it may be helpful to keep in mind specific cases you have worked with in guiding your answers. It is important that you do not share any identifiable information about these cases during the interview, however to consider how these may have impacted on you and how you may wish to approach the following questions.

About the team / context

1. Please can you tell me about the structure of the team you work in?
   - Prompting questions
     Do you have a lot of contact, or any shared spaces with your colleagues – what is the impact of this?
2. Please can you tell me how long you have been working in cleft settings and how many years of experience you have in your current role.
3. How many days do you work in cleft settings?
4. Please can you tell me which professionals are involved in the pre-orthognathic clinics and the extent of different professionals involvement?

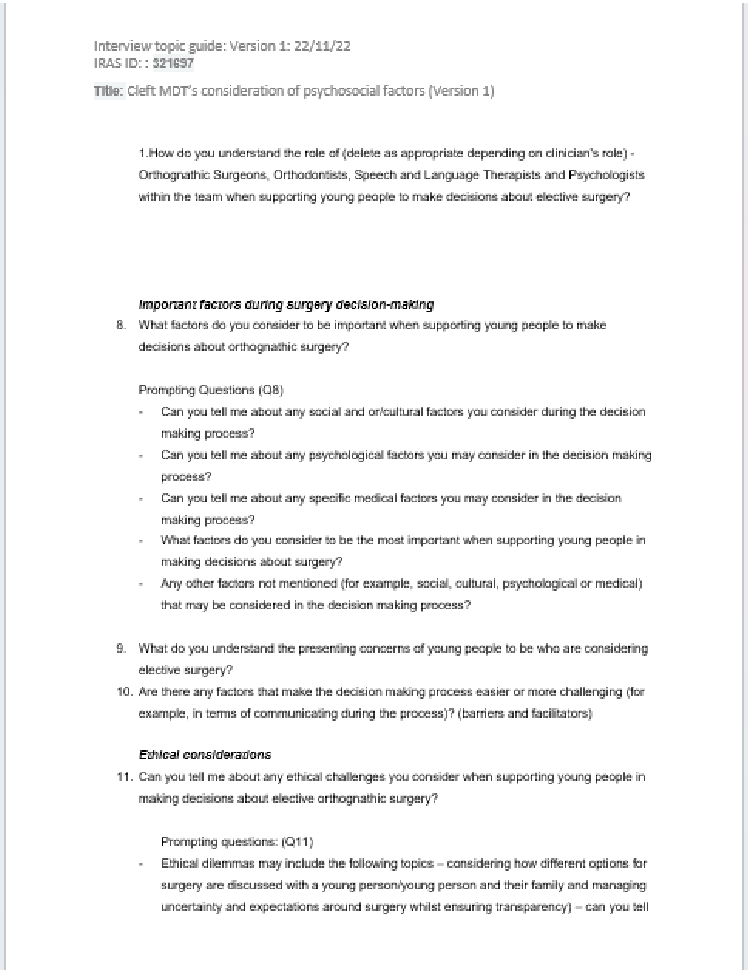
5. How do you experience communication and discussions within your team with regards to supporting young people to make decisions about surgery?

Prompting questions (Q5)
1.Can you tell me about the format of team meetings/discussions and how you experience your role within these.
2.Can you tell me about how your team comes together to make decisions – particularly in relation to decision making around Orthognathic Surgery.

Understanding roles

6. How do you understand your role in relation to supporting young people in making decisions about elective surgery?
7. How do you understand the role of other members of the team in supporting young people in making decisions about elective surgery?

Prompting questions: (Q7)



Interview topic guide: Version 1: 22/11/22
IRAS ID: : 321697

Title: Cleft MDT's consideration of psychosocial factors (Version 1)

1.How do you understand the role of (delete as appropriate depending on clinician's role) - Orthognathic Surgeons, Orthodontists, Speech and Language Therapists and Psychologists within the team when supporting young people to make decisions about elective surgery?

Important factors during surgery decision-making

8. What factors do you consider to be important when supporting young people to make decisions about orthognathic surgery?

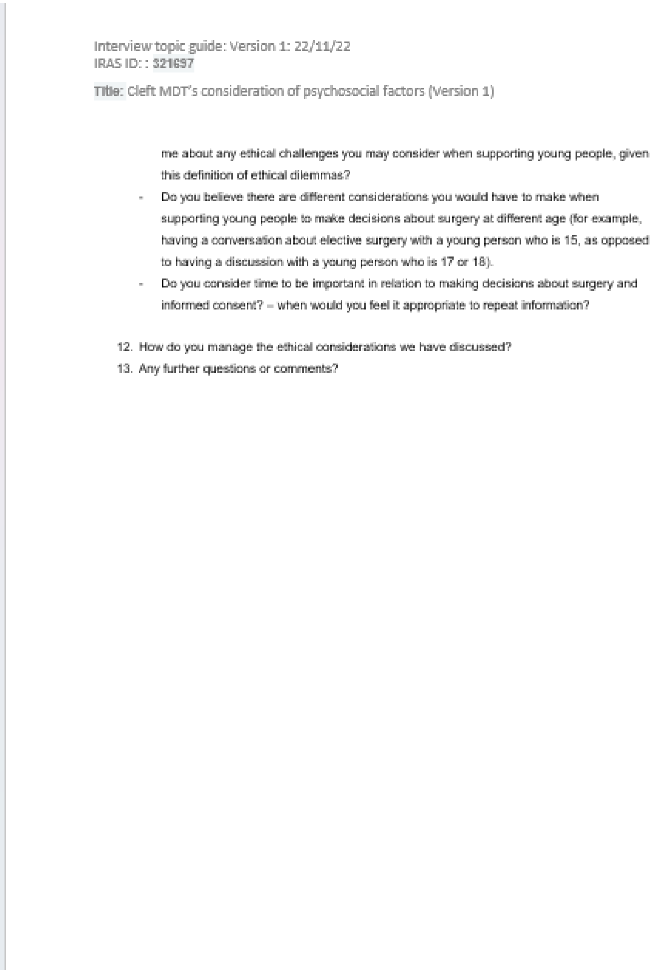
Prompting Questions (Q8)
- Can you tell me about any social and or/cultural factors you consider during the decision making process?
- Can you tell me about any psychological factors you may consider in the decision making process?
- Can you tell me about any specific medical factors you may consider in the decision making process?
- What factors do you consider to be the most important when supporting young people in making decisions about surgery?
- Any other factors not mentioned (for example, social, cultural, psychological or medical) that may be considered in the decision making process?

9. What do you understand the presenting concerns of young people to be who are considering elective surgery?
10. Are there any factors that make the decision making process easier or more challenging (for example, in terms of communicating during the process)? (barriers and facilitators)

Ethical considerations

11. Can you tell me about any ethical challenges you consider when supporting young people in making decisions about elective orthognathic surgery?

Prompting questions: (Q11)
- Ethical dilemmas may include the following topics – considering how different options for surgery are discussed with a young person/young person and their family and managing uncertainty and expectations around surgery whilst ensuring transparency) – can you tell



Interview topic guide: Version 1: 22/11/22
IRAS ID: : 321697

Title: Cleft MDT's consideration of psychosocial factors (Version 1)

me about any ethical challenges you may consider when supporting young people, given this definition of ethical dilemmas?
- Do you believe there are different considerations you would have to make when supporting young people to make decisions about surgery at different age (for example, having a conversation about elective surgery with a young person who is 15, as opposed to having a discussion with a young person who is 17 or 18).
- Do you consider time to be important in relation to making decisions about surgery and informed consent? – when would you feel it appropriate to repeat information?

12. How do you manage the ethical considerations we have discussed?
13. Any further questions or comments?

Appendix 13: Recruitment Email

Recruitment email to professionals (Version 1) – 22/11/22

Dear colleague,

My name is Holly Clegg and I am a Trainee Clinical Psychologist studying for the professional doctorate in Clinical Psychology at the University of East Anglia.

I am emailing to let you know about a new research project we are currently recruiting to looking at healthcare professionals understanding of psychosocial factors when supporting young people in making decisions about orthognathic surgery.

The study will involve taking part in an interview on Microsoft Teams, in which you will be asked some questions about the decision-making making process and the different factors you consider during these interactions.

We are currently looking to recruit Speech and Language Therapists, Orthognathic Surgeons and Orthodontists who have access to Microsoft Teams and are comfortable talking about their experiences. A participant information sheet has been attached with this email which provides further information about the study's aims and what would be involved if taking part.

As a thank you for taking part, clinicians will receive a £10 Amazon voucher.

Should you be interested in taking part in the study, or have any further questions, please do not hesitate to contact me on h.clegg@uea.ac.uk.

I would also be grateful if you could share this information with any other colleagues in your team who may not be aware of the project and may wish to take part.

Your support is much appreciated.

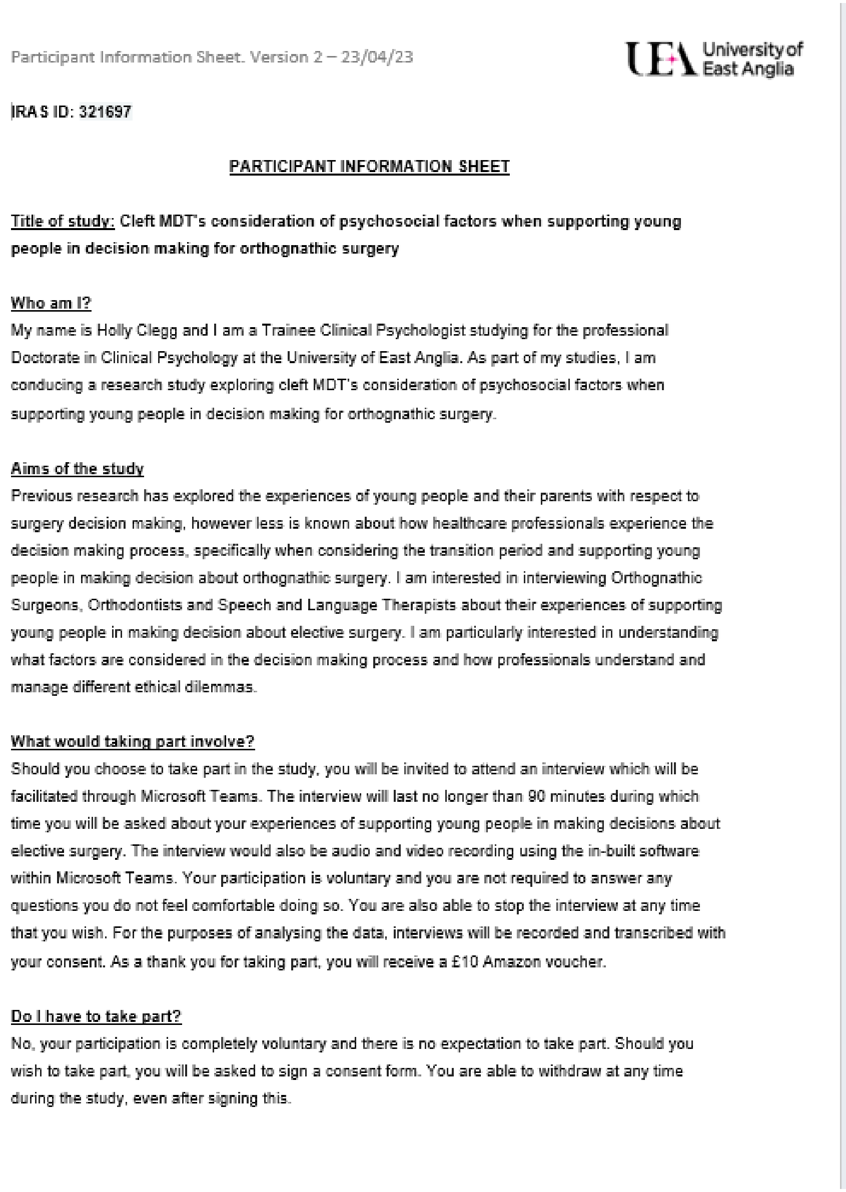
Yours sincerely,

Holly Clegg

Trainee Clinical Psychologist and Chief Investigator.

h.clegg@uea.ac.uk

Appendix 14. Participant Information Sheet



UEA University of East Anglia

IRAS ID: 321697

PARTICIPANT INFORMATION SHEET

Title of study: Cleft MDT's consideration of psychosocial factors when supporting young people in decision making for orthognathic surgery

Who am I?

My name is Holly Clegg and I am a Trainee Clinical Psychologist studying for the professional Doctorate in Clinical Psychology at the University of East Anglia. As part of my studies, I am conducing a research study exploring cleft MDT's consideration of psychosocial factors when supporting young people in decision making for orthognathic surgery.

Aims of the study

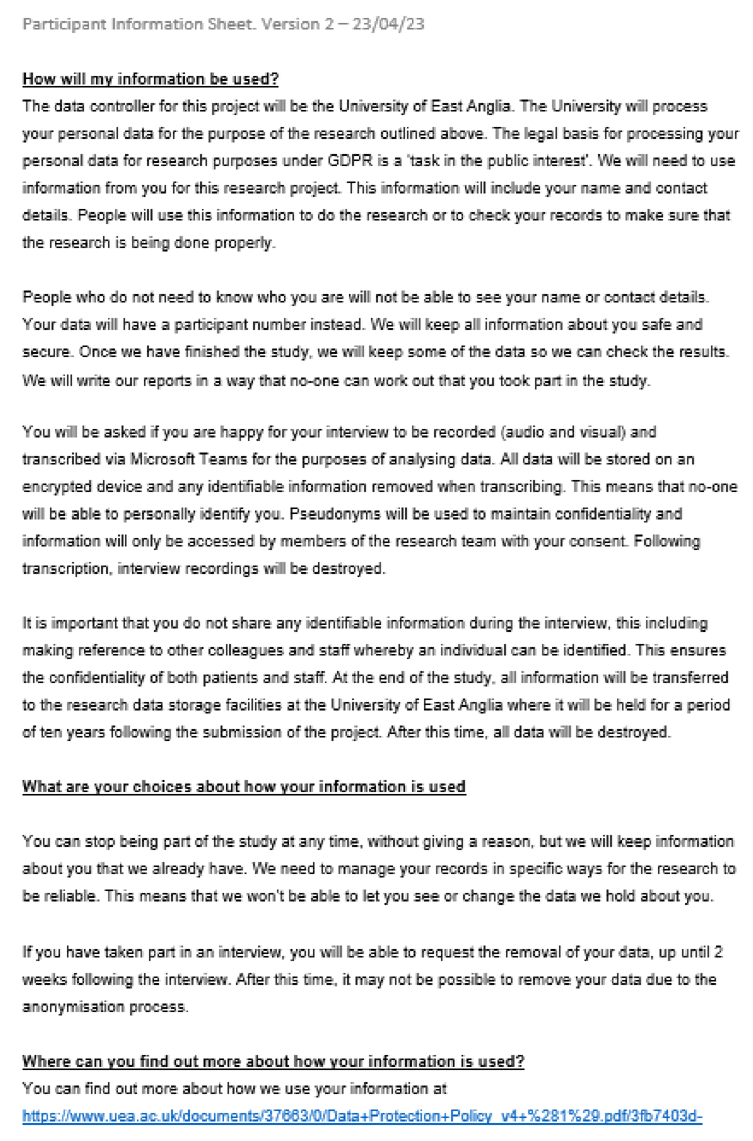
Previous research has explored the experiences of young people and their parents with respect to surgery decision making, however less is known about how healthcare professionals experience the decision making process, specifically when considering the transition period and supporting young people in making decision about orthognathic surgery. I am interested in interviewing Orthognathic Surgeons, Orthodontists and Speech and Language Therapists about their experiences of supporting young people in making decision about elective surgery. I am particularly interested in understanding what factors are considered in the decision making process and how professionals understand and manage different ethical dilemmas.

What would taking part involve?

Should you choose to take part in the study, you will be invited to attend an interview which will be facilitated through Microsoft Teams. The interview will last no longer than 90 minutes during which time you will be asked about your experiences of supporting young people in making decisions about elective surgery. The interview would also be audio and video recording using the in-built software within Microsoft Teams. Your participation is voluntary and you are not required to answer any questions you do not feel comfortable doing so. You are also able to stop the interview at any time that you wish. For the purposes of analysing the data, interviews will be recorded and transcribed with your consent. As a thank you for taking part, you will receive a £10 Amazon voucher.

Do I have to take part?

No, your participation is completely voluntary and there is no expectation to take part. Should you wish to take part, you will be asked to sign a consent form. You are able to withdraw at any time during the study, even after signing this.



How will my information be used?

The data controller for this project will be the University of East Anglia. The University will process your personal data for the purpose of the research outlined above. The legal basis for processing your personal data for research purposes under GDPR is a 'task in the public interest'. We will need to use information from you for this research project. This information will include your name and contact details. People will use this information to do the research or to check your records to make sure that the research is being done properly.

People who do not need to know who you are will not be able to see your name or contact details. Your data will have a participant number instead. We will keep all information about you safe and secure. Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

You will be asked if you are happy for your interview to be recorded (audio and visual) and transcribed via Microsoft Teams for the purposes of analysing data. All data will be stored on an encrypted device and any identifiable information removed when transcribing. This means that no-one will be able to personally identify you. Pseudonyms will be used to maintain confidentiality and information will only be accessed by members of the research team with your consent. Following transcription, interview recordings will be destroyed.

It is important that you do not share any identifiable information during the interview, this including making reference to other colleagues and staff whereby an individual can be identified. This ensures the confidentiality of both patients and staff. At the end of the study, all information will be transferred to the research data storage facilities at the University of East Anglia where it will be held for a period of ten years following the submission of the project. After this time, all data will be destroyed.

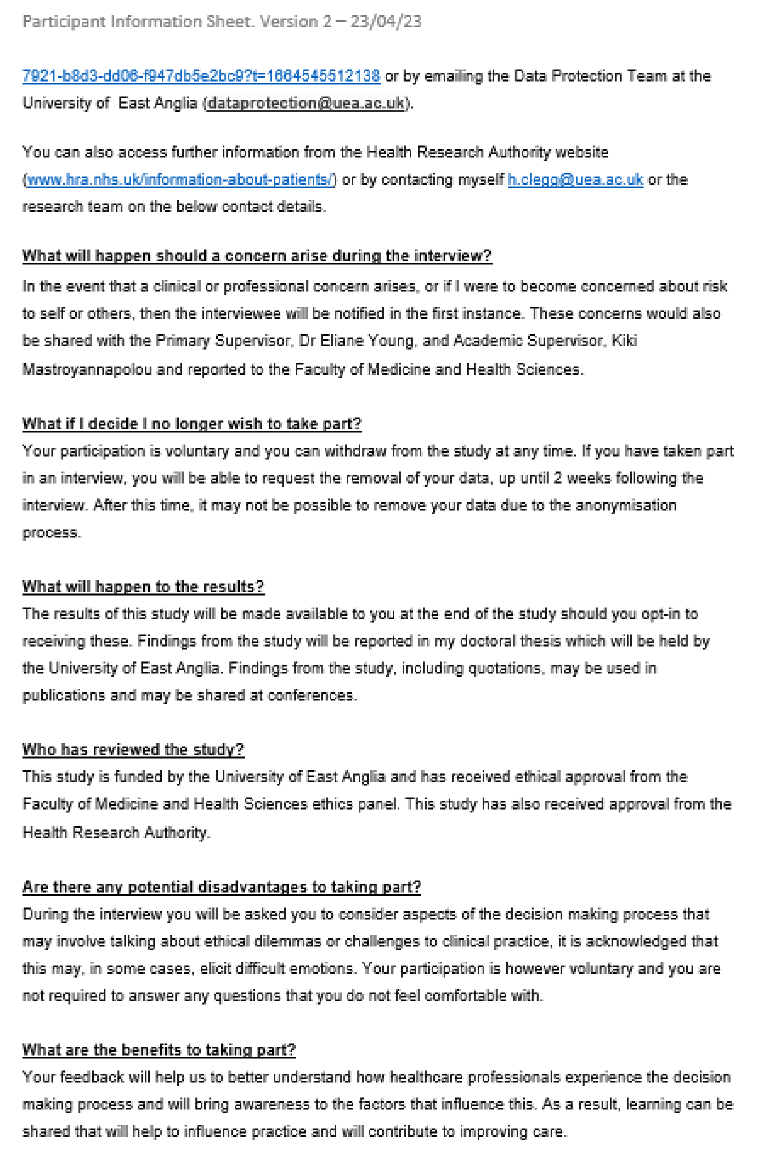
What are your choices about how your information is used

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have. We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

If you have taken part in an interview, you will be able to request the removal of your data, up until 2 weeks following the interview. After this time, it may not be possible to remove your data due to the anonymisation process.

Where can you find out more about how your information is used?

You can find out more about how we use your information at https://www.uea.ac.uk/documents/37663/0/Data+Protection+Policy_v4+%281%29.pdf/3fb7403d-



7921-b8d3-dd06-f947db5e2bc9?t=1664545512138 or by emailing the Data Protection Team at the University of East Anglia (dataprotection@uea.ac.uk).

You can also access further information from the Health Research Authority website (www.hra.nhs.uk/information-about-patients/) or by contacting myself h.clegg@uea.ac.uk or the research team on the below contact details.

What will happen should a concern arise during the interview?

In the event that a clinical or professional concern arises, or if I were to become concerned about risk to self or others, then the interviewee will be notified in the first instance. These concerns would also be shared with the Primary Supervisor, Dr Eliane Young, and Academic Supervisor, Kiki Mastroyannapolou and reported to the Faculty of Medicine and Health Sciences.

What if I decide I no longer wish to take part?

Your participation is voluntary and you can withdraw from the study at any time. If you have taken part in an interview, you will be able to request the removal of your data, up until 2 weeks following the interview. After this time, it may not be possible to remove your data due to the anonymisation process.

What will happen to the results?

The results of this study will be made available to you at the end of the study should you opt-in to receiving these. Findings from the study will be reported in my doctoral thesis which will be held by the University of East Anglia. Findings from the study, including quotations, may be used in publications and may be shared at conferences.

Who has reviewed the study?

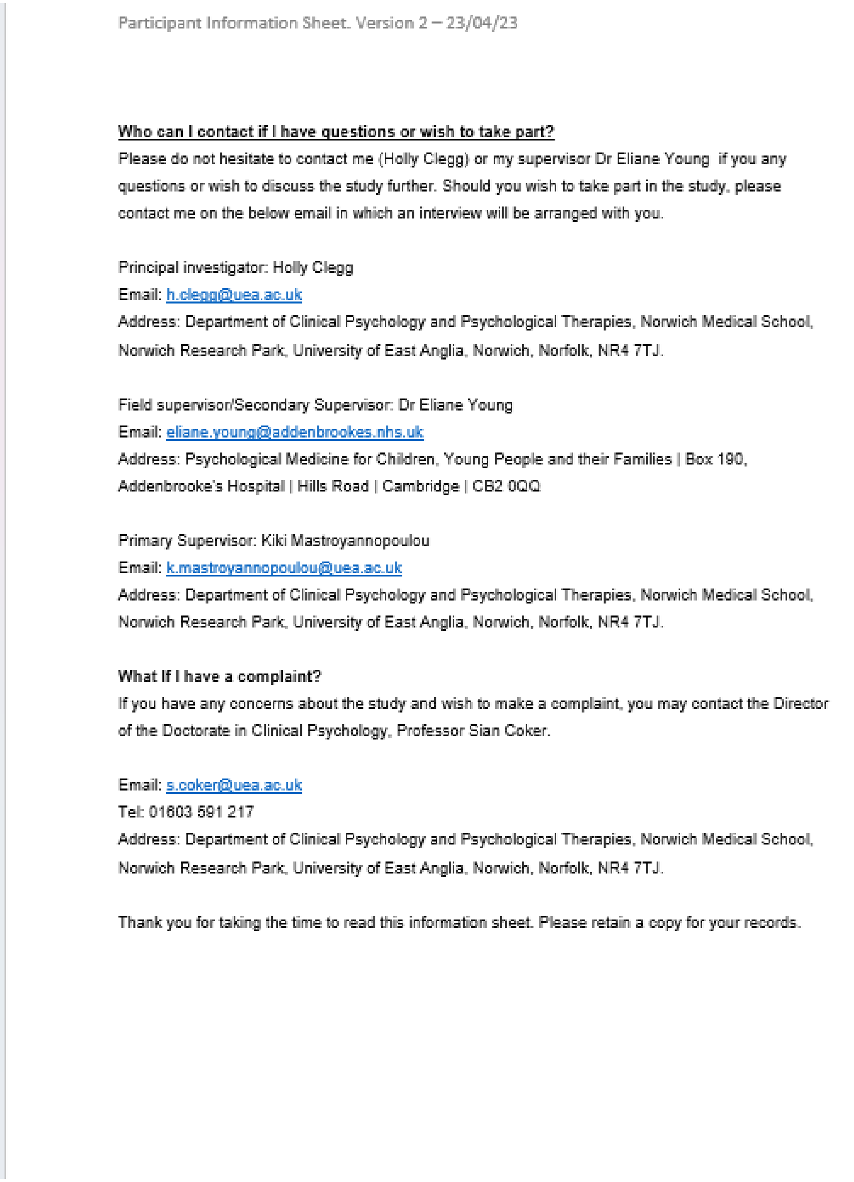
This study is funded by the University of East Anglia and has received ethical approval from the Faculty of Medicine and Health Sciences ethics panel. This study has also received approval from the Health Research Authority.

Are there any potential disadvantages to taking part?

During the interview you will be asked you to consider aspects of the decision making process that may involve talking about ethical dilemmas or challenges to clinical practice, it is acknowledged that this may, in some cases, elicit difficult emotions. Your participation is however voluntary and you are not required to answer any questions that you do not feel comfortable with.

What are the benefits to taking part?

Your feedback will help us to better understand how healthcare professionals experience the decision making process and will bring awareness to the factors that influence this. As a result, learning can be shared that will help to influence practice and will contribute to improving care.



Who can I contact if I have questions or wish to take part?

Please do not hesitate to contact me (Holly Clegg) or my supervisor Dr Eliane Young if you any questions or wish to discuss the study further. Should you wish to take part in the study, please contact me on the below email in which an interview will be arranged with you.

Principal investigator: Holly Clegg
Email: h.clegg@uea.ac.uk
Address: Department of Clinical Psychology and Psychological Therapies, Norwich Medical School, Norwich Research Park, University of East Anglia, Norwich, Norfolk, NR4 7TJ.

Field supervisor/Secondary Supervisor: Dr Eliane Young
Email: eliane.young@addenbrookes.nhs.uk
Address: Psychological Medicine for Children, Young People and their Families | Box 190, Addenbrooke's Hospital | Hills Road | Cambridge | CB2 0QQ

Primary Supervisor: Kiki Mastroyannopoulou
Email: k.mastroyannopoulou@uea.ac.uk
Address: Department of Clinical Psychology and Psychological Therapies, Norwich Medical School, Norwich Research Park, University of East Anglia, Norwich, Norfolk, NR4 7TJ.

What If I have a complaint?

If you have any concerns about the study and wish to make a complaint, you may contact the Director of the Doctorate in Clinical Psychology, Professor Sian Coker.

Email: s.coker@uea.ac.uk
Tel: 01603 591 217
Address: Department of Clinical Psychology and Psychological Therapies, Norwich Medical School, Norwich Research Park, University of East Anglia, Norwich, Norfolk, NR4 7TJ.

Thank you for taking the time to read this information sheet. Please retain a copy for your records.

Appendix 15: Example Coding framework

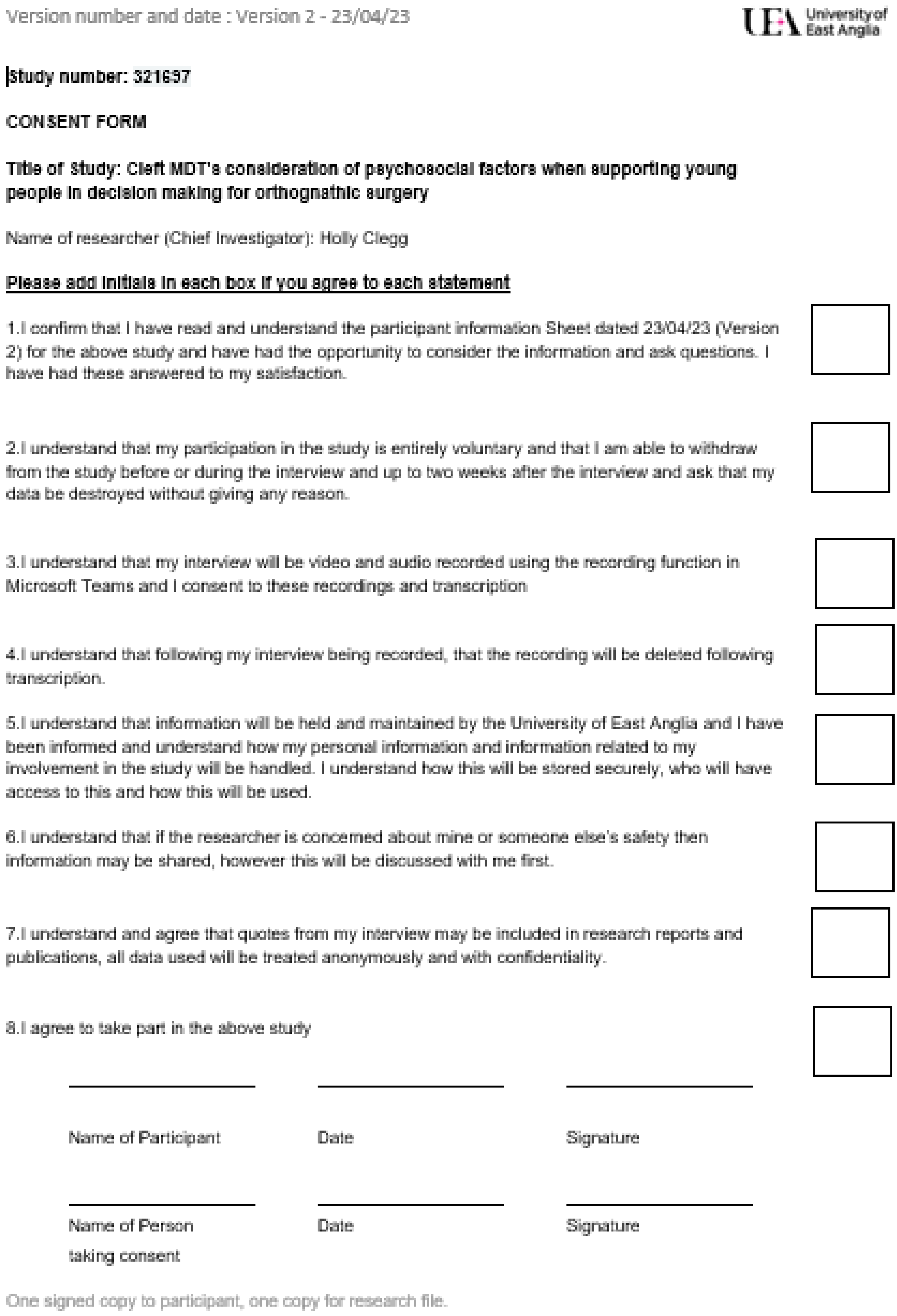
	Theme	**Definition**			
			Sub theme	**Definition**	**Example quotes**
1.	**Navigating the decision-making process**	**Factors that may facilitate or pose a barrier to collaboration and shared decision-making when discussing orthognathic surgery.**	**Finding a balance**	How clinicians maintain a balance between promoting patient autonomy and beneficence. Finding a position of 'compromise' in shared decision-making.	'I'm constantly challenged, because of the nature of our work. It's a constant challenge to try not to be to be paternalistic about things, but then there are sometimes when you just have to say no and so finding the right balance' **(P07)**
			External factors impacting on the decision-making process	Considering the role of social media and online communities on the decision-making process.	'Standard concerns about things being unmoderated, advice that we wouldn't necessarily recommend is being shared quite readily without anybody there to say, Oh no, no, don't do that.'**(P08)**
			Managing positions of power	Clinician awareness of power dynamics in the shared decision-making environment. Including positioning of the dental chair and number of professionals in one room.	'So even just the sort of power imbalance of that…of standing and him sitting.'(P09) 'and a dental chair which is behind the kind of half high wall so the patients don't walk on and see a dental chair, they come in and they seated area and we sit them down and we… we make sure it's that sort of...subtle things, but we make sure that we're always at eye level with them, so that the seats that the patient sit on are slightly higher than ours. So that when we are looking, we're on an equal level and we space it to make it sort of as informal as possible'.**(P03).**

			The process of eliciting motivations for surgery	Understanding what is important to the young person, and their motivations for surgery. Gender differences, and own position and influence in the SDM process is discussed.	‘under all that, there are reasons why people probably would like to have the benefits of having jaw surgery. But I think it's really important to kind of pick that out. You always want somebody…I mean, that's just how it was trained. You always wanted to get it out of the patient. Why they.. why they want surgery.’ **(P05).** ‘girls are often a lot better than the boys, much better... Yeah, it's a bit of a generalisation, anecdotal observation, but they tend to be a bit more... yeah, I'd like my nose doing. I know I want my nose doing, I want my lip doing, but I've been told my top jaw is in the wrong position, so let's investigate that. But ultimately, I want my nose doing... Yeah? whereas the boys will come in... they're a bit like, um yeah, I'm okay, or... there’s nothing you can do, sort of thing, so their starting point is quite different.’ **(P02)** ‘I'm really cognizant of not creating…that awareness that, oh, maybe I look different. Is there something wrong with me? So I can distinctly remember seeing a young girl, you know… in…and she had no issues with how she looked. She had brilliant confidence, et cetera, and I'm very cognizant that I don't want to

					create something in her mind.' **(P05)**
2.	**'Team-centric'**	**How different professionals view their role and the role of others in the team, reflecting on opportunities for liaison and discussion with other MDT colleagues.**	**Sub theme**	**Definition**	**Example quotes**
			Opportunities to foster team communication	**Opportunities (or desire) to liaise and have discussions with other colleagues in the MDT.**	'the key to a strong and functional team is that you do have the opportunities just to have corridor conversations and that you do bump into your colleagues.' (P06) 'So there isn't really an opportunity to chat. So yeah, we probably need more time in between them occasionally.' **(P09)**
			Valued contribution of MDT members	**How clinicians view and seek input from other colleagues in the MDT**	'I think we have quite an important role to play in, in, in different ways. So I think I think we have an important role to play clinically, in terms of our specialism and in terms of advising.' **(P01).** 'I'd very much hate to work in a team where, for example, the surgeon led everything and was, you know, was seen as the clinical lead when everybody's a clinical lead because everybody's part to the service and to the input to the care is important.' **(P06)**
3.	**Health inequalities impacting on access to cleft care**	**Clinicians consider financial, employment, education and childcare**	**Equity and barriers to accessing care**	**Factors that preclude, or pose a barrier to patients**	'I don't see the same mixture of backgrounds in our cleft orthognathic than I would expect

		issues that may preclude patients attending clinic. Consideration is also given to resources and professionals in attendance at each clinic.		**attending appointments for their cl/p.**	from kind of. It's in the general population now. That may be because there are so many other factors that preclude them actually turning up to clinic. You know, you think of all the hurdles that we put in place. They've got to be able to attend appointments, they've got to be able to have… they've got to be able to see a dentist on a regular basis to have good oral health. They've got to be able to engage with the communication from the hospital from the admin team, inviting them to appointments, and I would say all those things…exclude quite a significant proportion of people that we look after.' **(P03).**

Appendix 16. Participant Consent Form



Version number and date : Version 2 - 23/04/23

University of East Anglia

Study number: 321697

CONSENT FORM

Title of Study: Cleft MDT's consideration of psychosocial factors when supporting young people in decision making for orthognathic surgery

Name of researcher (Chief Investigator): Holly Clegg

Please add initials in each box if you agree to each statement

1.I confirm that I have read and understand the participant information Sheet dated 23/04/23 (Version 2) for the above study and have had the opportunity to consider the information and ask questions. I have had these answered to my satisfaction. ☐

2.I understand that my participation in the study is entirely voluntary and that I am able to withdraw from the study before or during the interview and up to two weeks after the interview and ask that my data be destroyed without giving any reason. ☐

3.I understand that my interview will be video and audio recorded using the recording function in Microsoft Teams and I consent to these recordings and transcription ☐

4.I understand that following my interview being recorded, that the recording will be deleted following transcription. ☐

5.I understand that information will be held and maintained by the University of East Anglia and I have been informed and understand how my personal information and information related to my involvement in the study will be handled. I understand how this will be stored securely, who will have access to this and how this will be used. ☐

6.I understand that if the researcher is concerned about mine or someone else's safety then information may be shared, however this will be discussed with me first. ☐

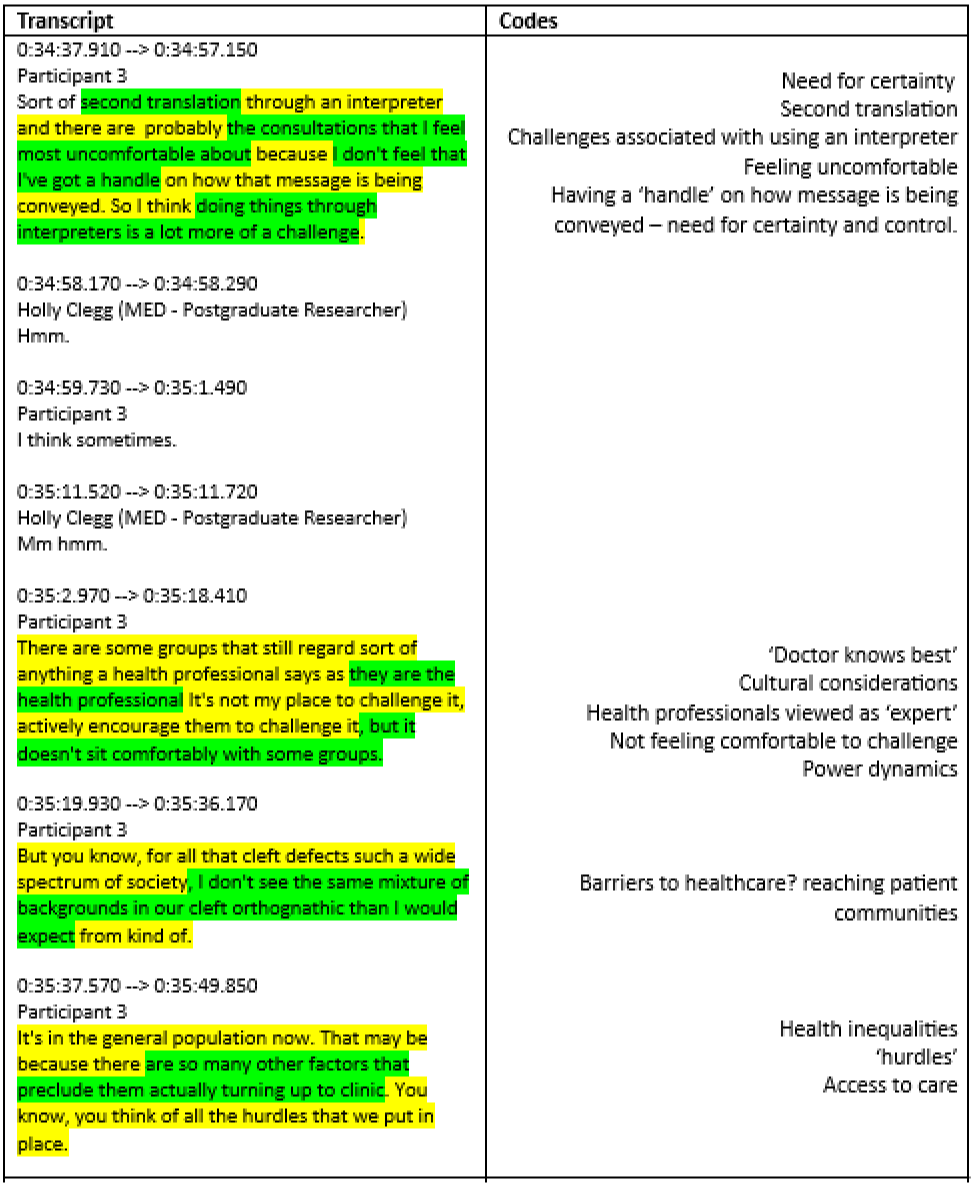
7.I understand and agree that quotes from my interview may be included in research reports and publications, all data used will be treated anonymously and with confidentiality. ☐

8.I agree to take part in the above study ☐

Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

One signed copy to participant, one copy for research file.

Appendix 17: Example coding of transcript

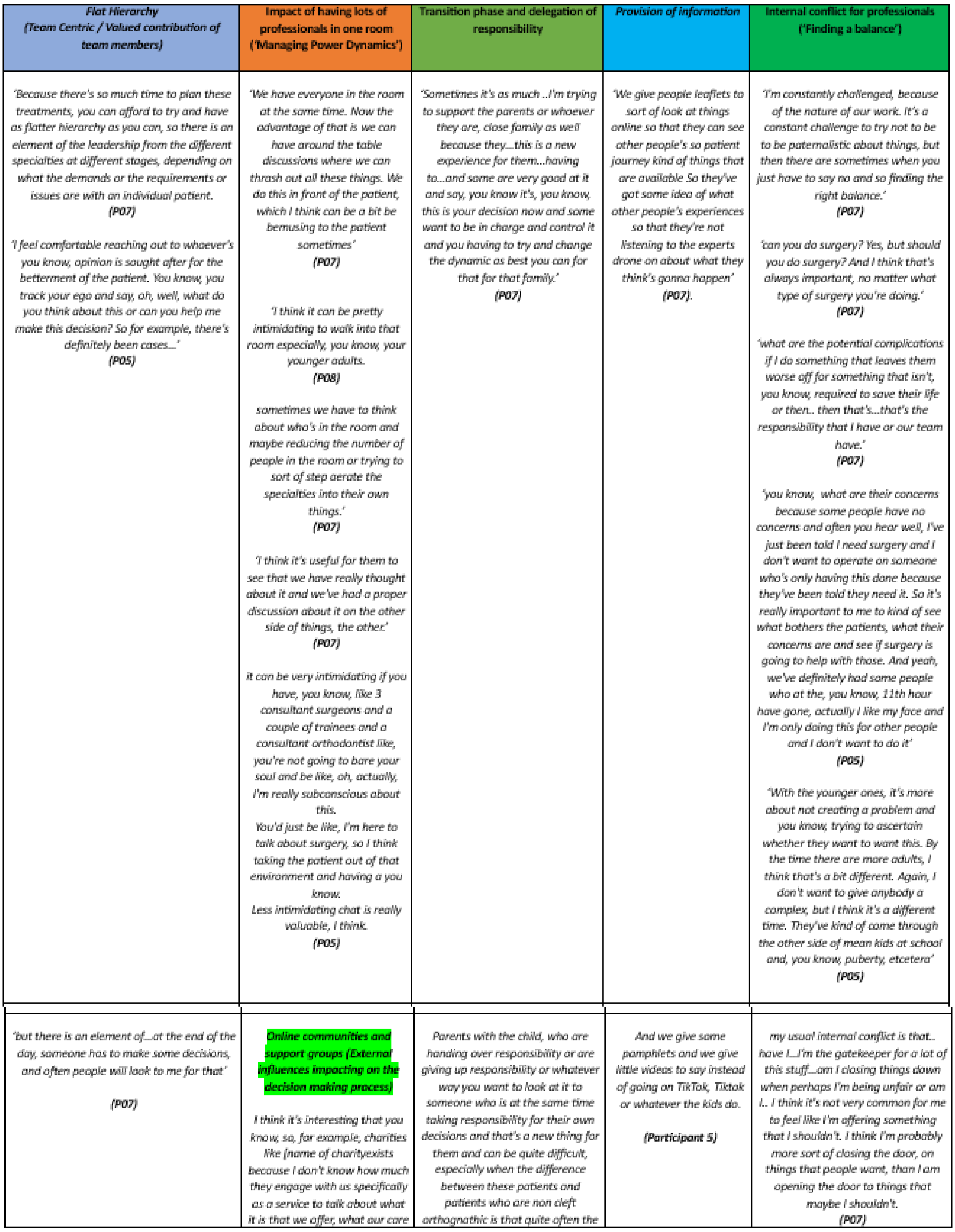


Transcript	Codes
0:34:37.910 --> 0:34:57.150 Participant 3 Sort of second translation through an interpreter and there are probably the consultations that I feel most uncomfortable about because I don't feel that I've got a handle on how that message is being conveyed. So I think doing things through interpreters is a lot more of a challenge.	Need for certainty Second translation Challenges associated with using an interpreter Feeling uncomfortable Having a 'handle' on how message is being conveyed – need for certainty and control.
0:34:58.170 --> 0:34:58.290 Holly Clegg (MED - Postgraduate Researcher) Hmm.	
0:34:59.730 --> 0:35:1.490 Participant 3 I think sometimes.	
0:35:11.520 --> 0:35:11.720 Holly Clegg (MED - Postgraduate Researcher) Mm hmm.	
0:35:2.970 --> 0:35:18.410 Participant 3 There are some groups that still regard sort of anything a health professional says as they are the health professional It's not my place to challenge it, actively encourage them to challenge it, but it doesn't sit comfortably with some groups.	'Doctor knows best' Cultural considerations Health professionals viewed as 'expert' Not feeling comfortable to challenge Power dynamics
0:35:19.930 --> 0:35:36.170 Participant 3 But you know, for all that cleft defects such a wide spectrum of society, I don't see the same mixture of backgrounds in our cleft orthognathic than I would expect from kind of.	Barriers to healthcare? reaching patient communities
0:35:37.570 --> 0:35:49.850 Participant 3 It's in the general population now. That may be because there are so many other factors that preclude them actually turning up to clinic. You know, you think of all the hurdles that we put in place.	Health inequalities 'hurdles' Access to care

Reflective journal entry

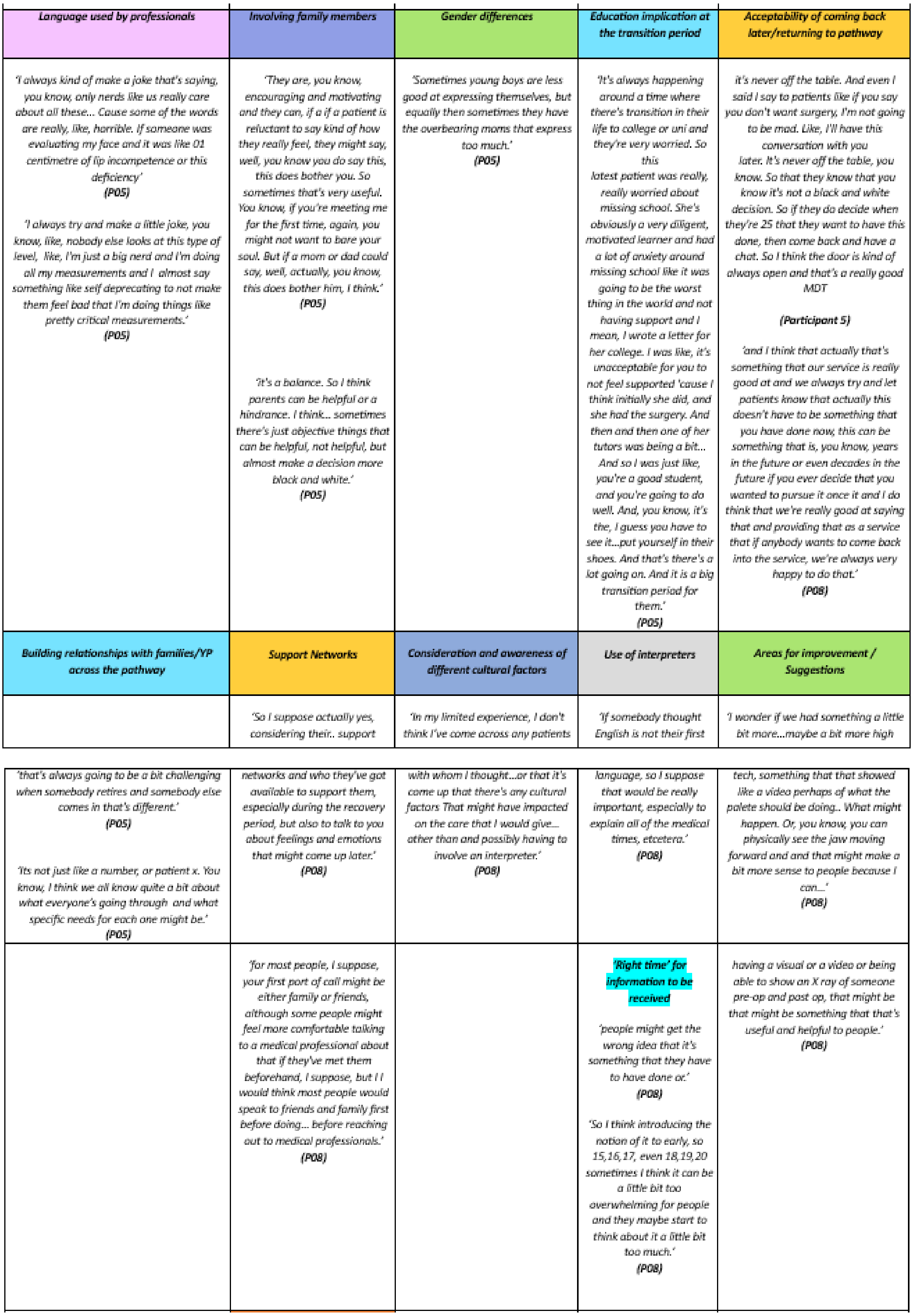
Clinicians are seeking a level of certainty in an environment whereby there is lot's of uncertainty to navigate, which is extremely difficult. As someone who takes an active role in EDI work, I was reassured to hear that clinicians are considering barriers to accessing healthcare, however at the same time I was also surprised to hear strong narratives about the health inequalities faced by different services, and the reported barriers for patients In accessing c

Appendix 18: Example theme development



Flat Hierarchy **(Team Centric / Valued contribution of team members)**	**Impact of having lots of professionals in one room ('Managing Power Dynamics')**	**Transition phase and delegation of responsibility**	**Provision of information**	**Internal conflict for professionals ('Finding a balance')**
'Because there's so much time to plan these treatments, you can afford to try and have as flatter hierarchy as you can, so there is an element of the leadership from the different specialties at different stages, depending on what the demands or the requirements or issues are with an individual patient. **(P07)** 'I feel comfortable reaching out to whoever's you know, opinion is sought after for the betterment of the patient. You know, you track your ego and say, oh, well, what do you think about this or can you help me make this decision? So for example, there's definitely been cases…' **(P05)**	'We have everyone in the room at the same time. Now the advantage of that is we can have around the table discussions where we can thrash out all these things. We do this in front of the patient, which I think can be a bit be bemusing to the patient sometimes' **(P07)** 'I think it can be pretty intimidating to walk into that room especially, you know, your younger adults. **(P08)** sometimes we have to think about who's in the room and maybe reducing the number of people in the room or trying to sort of step aerate the specialties into their own things.' **(P07)** 'I think it's useful for them to see that we have really thought about it and we've had a proper discussion about it on the other side of things, the other.' **(P07)** it can be very intimidating if you have, you know, like 3 consultant surgeons and a couple of trainees and a consultant orthodontist like, you're not going to bare your soul and be like, oh, actually, I'm really subconscious about this. You'd just be like, I'm here to talk about surgery, so I think taking the patient out of that environment and having a you know. Less intimidating chat is really valuable, I think. **(P05)**	'Sometimes it's as much ..I'm trying to support the parents or whoever they are, close family as well because they…this is a new experience for them…having to…and some are very good at it and say, you know it's, you know, this is your decision now and some want to be in charge and control it and you having to try and change the dynamic as best you can for that for that family.' **(P07)**	'We give people leaflets to sort of look at things online so that they can see other people's so patient journey kind of things that are available So they've got some idea of what other people's experiences so that they're not listening to the experts drone on about what they think's gonna happen' **(P07).**	'I'm constantly challenged, because of the nature of our work. It's a constant challenge to try not to be to be paternalistic about things, but then there are sometimes when you just have to say no and so finding the right balance.' **(P07)** 'can you do surgery? Yes, but should you do surgery? And I think that's always important, no matter what type of surgery you're doing.' **(P07)** 'what are the potential complications if I do something that leaves them worse off for something that isn't, you know, required to save their life or then.. then that's…that's the responsibility that I have or our team have.' **(P07)** 'you know, what are their concerns because some people have no concerns and often you hear well, I've just been told I need surgery and I don't want to operate on someone who's only having this done because they've been told they need it. So it's really important to me to kind of see what bothers the patients, what their concerns are and see if surgery is going to help with those. And yeah, we've definitely had some people who at the, you know, 11th hour have gone, actually I like my face and I'm only doing this for other people and I don't want to do it' **(P05)** 'With the younger ones, it's more about not creating a problem and you know, trying to ascertain whether they want to want this. By the time there are more adults, I think that's a bit different. Again, I don't want to give anybody a complex, but I think it's a different time. They've kind of come through the other side of mean kids at school and, you know, puberty, etcetera' **(P05)**
'but there is an element of…at the end of the day, someone has to make some decisions, and often people will look to me for that' **(P07)**	**Online communities and support groups (External influences impacting on the decision making process)** I think it's interesting that you know, so, for example, charities like [name of charityexists because I don't know how much they engage with us specifically as a service to talk about what it is that we offer, what our care	Parents with the child, who are handing over responsibility or are giving up responsibility or whatever way you want to look at it to someone who is at the same time taking responsibility for their own decisions and that's a new thing for them and can be quite difficult, especially when the difference between these patients and patients who are non cleft orthognathic is that quite often the	And we give some pamphlets and we give little videos to say instead of going on TikTok, Tiktok or whatever the kids do. **(Participant 5)**	my usual internal conflict is that.. have I…I'm the gatekeeper for a lot of this stuff…am I closing things down when perhaps I'm being unfair or am I.. I think it's not very common for me to feel like I'm offering something that I shouldn't. I think I'm probably more sort of closing the door, on things that people want, than I am opening the door to things that maybe I shouldn't. **(P07)**

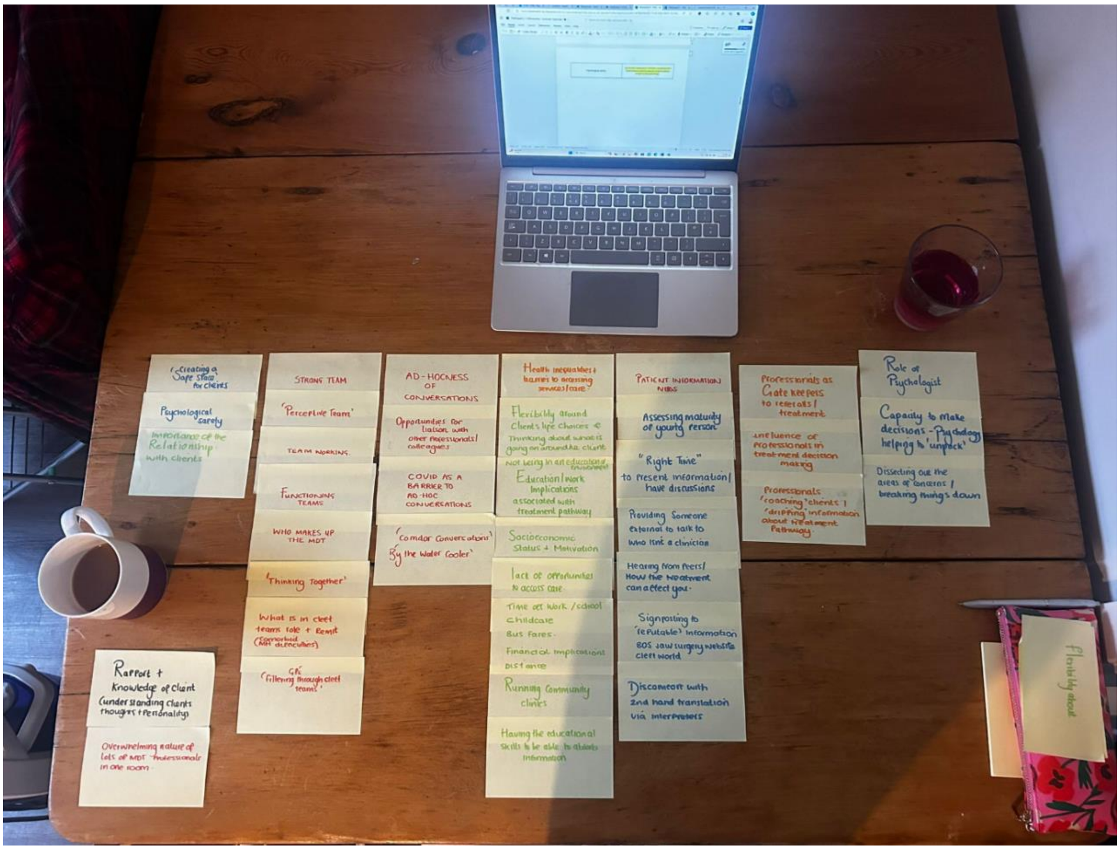
	pathways look like considering they're giving advice to families all over the country. **(P08)** *' I do belong to a couple of, umm, very general sort of cleft Facebook groups, and I think it's very helpful as a support network for families or just, you know, to ask a quick question or and but I think sometimes the fact that they're unmoderated and perhaps people sometimes giving advice that is is not what we might recommend as a as a medically based team or just thinking, for example, patients who are up and down the country receiving a different service and people saying.'* **(P08)** ***Health inequalities/ Equity of services*** *'So one appointment with everybody and then the whole MDT meeting, whereas we don't and whereas for three year check we would just do one MDT meeting everybody together and we would see* them separately if we *needed to. So just and then people being very shocked to the point of outrage that some people receiving this type of service and some people were not.'* **(P08)** *'standard concerns about things being unmoderated, advice that we wouldn't necessarily recommend is being shared quite readily without anybody there to say, Oh no, no, don't do that.'* **(P08)** *'And so they just kind of offloaded in this group and it's sort of hard to read because you can't then as a medical professional contact that parent and say, ahh, I heard you were unhappy about this. Sorry about that, because that has to be those boundaries of if they want to speak to us, to complain about it, that's a different matter.'* **(P08)**	*non cleft orthognathic patients, it's the first time they've possibly even set foot in the hospital and they've come because they have a specific issue that they want to address and they got an idea of what they want, whereas for the cleft patients, they've had all the decisions made for them up to that time.* **(Participant 7)**		*'when you're seeing kids very early on, you don't wanna create, you know, just because I think somebody would really benefit from jaw surgery, they might not...care about the things that I think will benefit.. do you know what I mean and it's even in'* **(P05)** *'So that's in the back of the mind, am I creating a disaster? am I gonna create a disaster here?'* **(P07)** *'it's not my own ego, but it is hard because you're like, have I done the right thing and operating on her and I, you know, she has good results.'* **(P05)** *' It's tough even psychologically for me to have operated.'* **(P05)** *'I'm really leery of saying anything too soon, I think.'* **(P05)**
Importance of Time	***Desire for more opportunities for discussion with colleagues***	***Being 'told' to have surgery, paternalism ('Finding a balance')***	***Eliciting motivations for surgery***	***Professional influence on decision making ('Finding a balance')***
'parking people with their thoughts for a while is probably my most commonly used tactic... if I feel either, the patient needs a bit more time to think.. to digest things....or if I need....if I'm not happy and I think that the possibly someone's trying to get something that is maybe not going to work for them' **(P07)**	*'I should make more time to try and set up a way of discussing people.... patients... difficult patients with other colleagues because it means... because of the very specialized nature of it....you have to go out of region to find someone who really knows what you're talking about...* *So that's ...that's one of the things that that I would like to develop and it's one of those on my list of all my a do list...'* **(P07)**	*'I think effectively we just kind of picked it apart a bit and I really recognised that this was somebody who was basically told this is the path and that's why he was having surgery and he's the one who said I like my face. I don't want it to change. I don't he.. he had some background anxiety full stop about having general anaesthetic and all these things that were going into. But I think I spent a lot of time in the end lot of chatting with the Clinical psychologist and whatnot trying to say like...Where is this coming from? Does he really want surgery and we're back and forth. He deferred the decision and ultimately, we had a really, like lengthy conversation at.. with multiple members of the team and ultimately he, he said no to surgery.'* **(P05)**	*'they're on this path and they're like this is the next task and I'm here to do it and I think under all that, there are reasons why people probably would like to have the benefits of having jaw surgery. But I think it's really important to kind of pick that out. You always want somebody....I mean, that's just how it was trained. You always wanted to get it out of the patient. Why they... why they want surgery.'* **(P05)**	*'I'm really cognizant of not creating...that awareness that, oh, maybe I look different. Is there something wrong with me? So I can distinctly remember seeing a young girl, you know, 13, and she had no issues with how she looked. She had brilliant confidence, et cetera, and I'm very cognizant that I don't want to create something in her mind'* **(P05)**
		'It was really paternalistic, so talking from behind the patient in the corner, basically telling them what was happening.' **(P05)**		

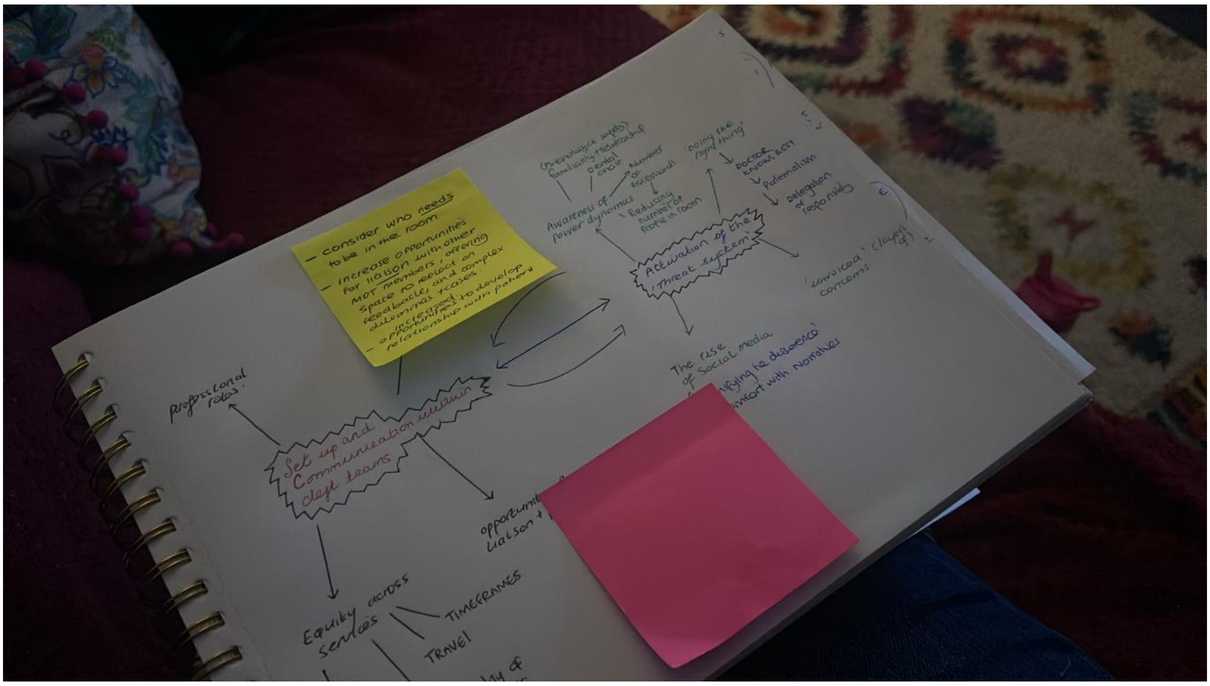


Language used by professionals	***Involving family members***	***Gender differences***	***Education implication at the transition period***	***Acceptability of coming back later/returning to pathway***
'I always kind of make a joke that's saying, you know, only nerds like us really care about all these… Cause some of the words are really, like, horrible. If someone was evaluating my face and it was like 01 centimetre of lip incompetence or this deficiency' ***(P05)*** *'I always try and make a little joke, you know, like, nobody else looks at this type of level, like, I'm just a big nerd and I'm doing all my measurements and I almost say something like self deprecating to not make them feel bad that I'm doing things like pretty critical measurements.'* ***(P05)***	*'They are, you know, encouraging and motivating and they can, if a if a patient is reluctant to say kind of how they really feel, they might say, well, you know you do say this, this does bother you. So sometimes that's very useful. You know, if you're meeting me for the first time, again, you might not want to bare your soul. But if a mom or dad could say, well, actually, you know, this does bother him, I think.'* ***(P05)*** *'It's a balance. So I think parents can be helpful or a hindrance. I think… sometimes there's just objective things that can be helpful, not helpful, but almost make a decision more black and white.'* ***(P05)***	*'Sometimes young boys are less good at expressing themselves, but equally then sometimes they have the overbearing moms that express too much.'* ***(P05)***	*'It's always happening around a time where there's transition in their life to college or uni and they're very worried. So this* *latest patient was really, really worried about missing school. She's obviously a very diligent, motivated learner and had a lot of anxiety around missing school like it was going to be the worst thing in the world and not having support and I mean, I wrote a letter for her college. I was like, it's unacceptable for you to not feel supported 'cause I think initially she did, and she had the surgery. And then and then one of her tutors was being a bit… And so I was just like, you're a good student, and you're going to do well. And, you know, it's the, I guess you have to see it…put yourself in their shoes. And that's there's a lot going on. And it is a big transition period for them.'* ***(P05)***	*it's never off the table. And even I said I say to patients like if you say you don't want surgery, I'm not going to be mad. Like, I'll have this conversation with you* *later. It's never off the table, you know. So that they know that you know it's not a black and white decision. So if they do decide when they're 25 that they want to have this done, then come back and have a chat. So I think the door is kind of always open and that's a really good MDT* ***(Participant 5)*** *'and I think that actually that's something that our service is really good at and we always try and let patients know that actually this doesn't have to be something that you have done now, this can be something that is, you know, years in the future or even decades in the future if you ever decide that you wanted to pursue it once it and I do think that we're really good at saying that and providing that as a service that if anybody wants to come back into the service, we're always very happy to do that.'* ***(P08)***
Building relationships with families/YP across the pathway	***Support Networks***	***Consideration and awareness of different cultural factors***	***Use of interpreters***	***Areas for improvement / Suggestions***
'that's always going to be a bit challenging when somebody retires and somebody else comes in that's different.' ***(P05)*** *'Its not just like a number, or patient x. You know, I think we all know quite a bit about what everyone's going through and what specific needs for each one might be.'* ***(P05)***	*'So I suppose actually yes, considering their.. support networks and who they've got available to support them, especially during the recovery period, but also to talk to you about feelings and emotions that might come up later.'* ***(P08)***	*'In my limited experience, I don't think I've come across any patients with whom I thought…or that it's come up that there's any cultural factors That might have impacted on the care that I would give… other than and possibly having to involve an interpreter.'* ***(P08)***	*'If somebody thought English is not their first language, so I suppose that would be really important, especially to explain all of the medical times, etcetera.'* ***(P08)***	*'I wonder if we had something a little bit more…maybe a bit more high tech, something that that showed like a video perhaps of what the palete should be doing.. What might happen. Or, you know, you can physically see the jaw moving forward and and that might make a bit more sense to people because I can…'* ***(P08)***
	'for most people, I suppose, your first port of call might be either family or friends, although some people might feel more comfortable talking to a medical professional about that if they've met them beforehand, I suppose, but I I would think most people would speak to friends and family first before doing… before reaching out to medical professionals.' ***(P08)***		***'Right time' for information to be received*** *'people might get the wrong idea that it's something that they have to have done or.'* ***(P08)*** *'So I think introducing the notion of it to early, so 15,16,17, even 18,19,20 sometimes I think it can be a little bit too overwhelming for people and they maybe start to think about it a little bit too much.'* ***(P08)***	*having a visual or a video or being able to show an X ray of someone pre-op and post op, that might be that might be something that that's useful and helpful to people.'* ***(P08)***

	Facial aesthetics			
'So somebody perhaps who has developmental language disorder or you know just has those and any associated language difficulties or developmental delays or.learning needs' **(P08)**	*'It's probably based on how, on their facial appearance and and they.. want that to, you know, improve or change'* **(P08)** *'And I think that I think that that's a big factor for quite a lot of patients in, in terms of going forward that actually that it will have such an impact on the way that they they look and how they and and thus possibly how they, how they feel about feel about themselves facially and and and appearance wise and thinking about how anybody might not want to go forward.'* **(P08)** *'Some people, I think, don't don't don't want their.. don't want their profile to change, don't want their face to change and and that can be become quite a a big issue for them so might not want to go forward because of that'* **(P08)**			

Appendix 19: Visual representation of searching for themes and subthemes




- consider who needs to be in the room
- increase opportunities for liaison with other MDT members, offering space to reflect on feedback, and complex dilemmas + cases
- increased opportunities to develop relationship with patient
(Psychological safety) familiarity + relationship
Dental chair
Number of professionals
Reducing number of people in room
Awareness of power dynamics
'Doing the right thing'
DOCTOR KNOWS BEST
Paternalism
Delegation of responsibility
Activation of the 'Threat system'
'unvoiced' concerns
The rise of Social media
professional roles
Set up and Communication within dept teams
opportunities liaison +
Equity across services
TIMEFRAMES
TRAVEL

Appendix 20: **Worked example of reviewing and defining themes**

Main Theme	Sub Themes
Theme 1: Impact of the MDT Environment	• *Challenges associated with the MDT coming together with the patient in one room* • *Advantages of meeting as an MDT* • *Creating psychological safety in the MDT environment*
Theme 2: 'Digging deep' around the unvoiced	• *Clinician skill in exploring patient concerns* • *Perceived unacceptability of asking for surgery for cosmetic/aesthetic reasons* • *Gender differences* • *The role of Psychology in the Cleft MDT* • *Responsibility for mental health*
Theme 3: Influence of systems on decision-making	• *Choice of words and language used during SDM* • *Acknowledgement and influence of paternalism* • *Influence of support networks* • *Consistency in the level of detailed information provided to patients*
Theme 4: Awareness of Health Inequalities and barriers to accessing cleft care.	• *Factors that exclude* • *Financial and travel implications* • *Equity across services.*
Theme 5: The rise of social media and online platforms	• *Magnifying the difference* • *Influence of patient/peer support groups and communities* • *Signposting*
Theme 6: Internal conflict experienced by professionals	• *Autonomy vs beneficence* • *'Doing the right thing'*
Theme 7: Considering culture and religion with SDM.	• *'Doctor knows best'* • *Religious orientation and associated ethical dilemmas*
Theme 8: Acknowledging the gravity of the decision being made	• Commitment to the treatment pathway

	• Delegating responsibility at the transition period

Table of themes draft 2

Themes and subthemes	
Theme	**Subthemes**
Theme 1.: Activation of the 'threat' system	• *''Doing the right thing'* • *Rise of social media* • *Awareness of power dynamics* • *"unvoiced" layers of concern*
Theme 2: 'Team-centric" – Implementation of the 20 year pathway.	• *Opportunities for liaison* • *Equity and health inequalities* • *Perception of professional roles*

Final themes

Themes and subthemes	
Theme	**Subthemes**
Theme 1: Navigating the decision-making process	• *Finding a 'balance'* • *External factors impacting on the decision-making process* • *Managing positions of power* • *The process of eliciting motivations for surgery*
Theme 2: 'Team-centric"	• *Opportunities to foster team communication* • *Valued contribution of MDT members*
Theme 3: Health Inequalities impacting on access to cleft care.	• *Equity and barriers to accessing care*