



End of pathway cleft surgery: Exploring the patient-reported outcomes and
young people's decision making experiences.

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Table of Contents

Acknowledgements	5
Thesis Portfolio Abstract	6
Chapter One: Setting the scene	8
Chapter Two: Systematic Review	21
Chapter Three: Empirical paper	73
Chapter Four: Extended methodology	112
Chapter Five: Extended results and discussion.....	127
References	145
List of appendices	172
Appendix 1. The Cleft Palate-Craniofacial Journal Manuscript Preparation Guidelines.....	173
Appendix 2. Data extraction template.....	181
Appendix 3. Quality criteria for critical appraisal of included studies	182
Appendix 4. Quality criteria rating table with inter-rater checks	185
Appendix 5. Table of excluded studies with reasons	187
Appendix 6. Health Research Authority (HRA) ethical approval letter	188

Appendix 7. Interview guide 196

Appendix 8. Patient invitation letter 199

Appendix 9. Recruitment advert 201

Appendix 10. Participant information sheet 202

Appendix 11. Participant consent form 207

Appendix 12. Demographic information form 209

Appendix 13. Example of coded transcript extract 211

Appendix 14. Example coding and theme development 214

Appendix 15. Supplementary participant quotes to support derived themes 216

List of tables and figures

Table 1. Level of evidence for PROs by study design 31

Table 2. Overall quality ratings for included studies based on PROs 33

Table 3. Patient-reported outcome measures used 34

Table 4. Study characteristics and summary of key findings from included studies 37

Table 5. Demographic information for study participants 81

Table 6. Themes and subthemes 83

Figure 1. Key surgical and therapeutic interventions for CL/P patients across the
lifespan 10

Figure 2. Outcome of search process.....30

Figure 3. Thematic diagram83

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Thesis Portfolio Abstract

Objective: Literature exploring how young people (YP) experience cleft surgery at the end of the treatment pathway is limited, both in terms of their reported outcomes and their experience of deciding whether to undergo surgery. This thesis aimed to add to the cleft field by reviewing the patient-reported outcomes (PROs) of end of pathway cleft surgery and exploring YP's experiences of deciding whether to undergo orthognathic surgery (OS; an end of pathway cleft surgery).

Design: A systematic literature search identified studies measuring the PROs of undergoing end of pathway cleft surgery. To explore OS decision making experiences a qualitative design was employed and interviews conducted with twelve YP.

Results: The 22 studies measuring PROs varied in methodological quality; most were small scale and none utilised a measure validated in the cleft population, meaning it is hard to draw conclusions about end of pathway cleft surgery from the patient perspective. Thematic Analysis of YP's accounts resulted in the development of four themes to depict YP's decision making experiences: 1) *Awareness of difference*, 2) *Committing to the process*, 3) *Others facilitating decision making* and 4) *Responsibility on my shoulders*.

Conclusions: This thesis reveals the difficulty in determining PROs of end of pathway cleft surgery due to the methodological challenges and the heterogeneity of what, how and when outcomes are measured. It demonstrates the contextual, social and personal complexities YP experienced in the process of deciding about OS during a period of developmental transition. Theoretical, clinical and research implications are discussed.

Key words: cleft, surgery, patient-reported outcomes, decision making,
young person, orthognathic

Chapter One: Setting the scene

Cleft lip and or palate

Cleft lip and or palate (CL/P) is the most common craniofacial anomaly. Although estimates vary, globally and in the United Kingdom (UK) approximately 1 in 700 babies are born with a CL/P (Goodacre & Swan, 2008; Mossey & Castilla, 2001). A CL/P occurs when separate areas of the face do not join together properly during early pregnancy, leaving a gap (cleft) in the upper lip, palate, or both. Clefts can manifest unilaterally (on one side) or bilaterally (on both sides of the face) and also vary in severity. The causes of non-syndromic CL/P's are largely unknown, and a proportion of syndromic CL/P's are part of a genetic syndrome.

Treatment pathway and aims

Management of patients with CL/P is complex. In the National Health Service (NHS) treatment follows a structured 20-year care pathway that is tailored to the individual depending on cleft type and severity (Colbert, Green, Brennan, & Mercer, 2015; NHS England, 2013). Typically beginning before birth and extending throughout development into early adulthood, treatment focuses upon improving quality of life by addressing the functional (feeding, hearing, speech) and appearance related (dentition, facial structure/features) consequences of the CL/P.

Surgical intervention forms a major part of treatment with multiple surgeries across the lifespan (for an overview see Goodacre & Swan, 2008). Surgery begins with the initial repair of the cleft lip and or palate in the first year of life followed by further clinically recommended surgeries until the end of the treatment pathway, when patients and families are offered various elective surgeries to further improve function and alter appearance. For example, as typical growth of the jaw and midface

can be impeded by the unavoidable scarring from primary cleft surgery (Andersen, Nørholt, Kùseler, Jensen, & Pedersen, 2012; Shetye, 2004) patients may be offered surgery to alter jaw alignment (Orthognathic Surgery, OS or Distraction Osteogenesis, DO), to alter the appearance and function of the nose (Rhinoplasty), revise lip scarring (Lip Revision) or minimise hyper-nasal speech (Secondary Speech Surgery). In addition to surgical, dental and orthodontic input specialist therapeutic interventions including Speech and Language Therapy and Clinical Psychology are available across the pathway (see Figure 1). The overarching aim of this complex care pathway is to offer treatment that minimises the adverse consequences of the CL/P to ensure children, young people and adults are not disadvantaged and can reach their full potential (Goodacre & Swan, 2008; Mossey, Little, Munger, Dixon, & Shaw, 2009; Robin et al., 2006). As such all CL/P outcomes fall within domain 2 of the NHS Outcomes Framework ‘Enhancing quality of life for people with long-term conditions’ (Department of Health, 2016).

Although there is a defined pathway for cleft care, services operate across the lifespan as adults of any age can access further cleft treatment, if desired. Due to the variety of functions affected by CL/P across the course of development, such as speech, hearing, teeth, appearance and psychosocial wellbeing, treatment for CL/P in developed countries is delivered by specialist multidisciplinary teams with expertise in each domain (Hodgkinson et al., 2005). Since the Clinical Standards Advisory Group (CSAG) report and subsequent reorganization of services two decades ago (Sandy et al., 1998), the provision of Clinical Psychology has become part of the multidisciplinary approach to cleft care. Psychologists are well-placed to help patients and families adjust to living with a CL/P and its associated treatment, including screening for emotional distress and offering support and intervention

across the pathway. It has been suggested that in addition to minimising the psychosocial consequences of being born with and treated for CL/P, psychologists might also contribute to reducing the number of secondary procedures that are governed by patient choice (Goodacre & Swan, 2008). In line with best practice it is recommended that where OS is clinically indicated patients are invited to attend a pre-orthognathic clinic to undergo a multi-disciplinary team assessment. In addition to meeting with the surgical team, patients have a speech and language assessment and a psychological consultation where patients complete questionnaires to assess satisfaction with appearance, psychological adjustment, and expectations for surgery. Further sessions with the psychologist are available should concerns arise or should the YP require further support with decision making.

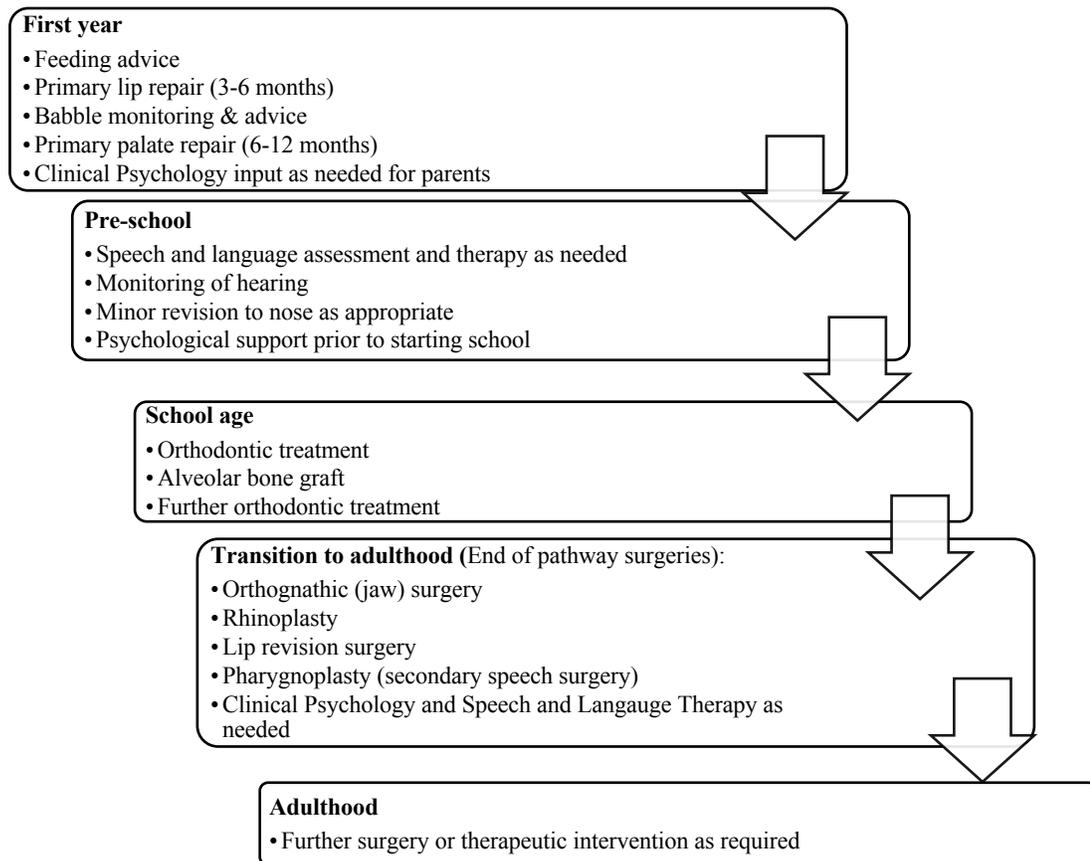


Figure 1. Key surgical and therapeutic interventions for CL/P patients across the lifespan.

Psychosocial sequelae of living with and being treated for a CL/P

The face forms an important part of self-identity and also plays a central role in social interactions including how we might be perceived by others (Strauss et al., 2007). Given that oral clefts affect facial appearance and function CL/P and its treatment can create multiple challenges and burdens for patients and families, with the potential to affect psychological and social wellbeing. The literature exploring patients and families experiences of living with a CL/P highlights both the negative and positive impact of CL/P on psychosocial functioning and quality of life. Some of the challenges encountered by patients with CL/P may include difficulties in social functioning, dissatisfaction with appearance, coping with the burden of ongoing treatment, in addition to the impact of the condition and its treatment on psychological wellbeing.

In particular, people with a CL/P appear vulnerable to stigma and teasing with consistent reports from patients and families across studies of teasing related to the CL/P (Alansari, Bedos, & Allison, 2014; Hunt, Burden, Hepper, Stevenson, & Johnston, 2007; Stock, Feragen, & Rumsey, 2016; Tiemens, Nicholas, & Forrest, 2013; Turner, Rumsey, & Sandy, 1998; Turner, Thomas, Dowell, & Rumsey, 1997). The literature suggests teasing to be more prevalent in those with CL/P compared to non-affected peers (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Hunt et al., 2007), although the experience of teasing itself (rather than the CL/P) was found to be a significant predictor of psychosocial distress (Hunt et al., 2006). It is suggested that the impact of teasing could depend upon the degree to which individuals internalise the stigma as in a recent qualitative study, where a majority of participants reported experiences of teasing, some felt the effects of teasing were lasting while others did not (Stock et al., 2016). Such variation in emotional

resilience could be explained by various protective factors such as having friendships which support positive self-perception (Feragen, Kvalem, Rumsey, & Borge, 2010).

Studies also highlight how the experience of living with a CL/P can change over time. Adults in a qualitative study reported feeling stigmatised, different from others and burdened by the treatment pathway in their childhood and adolescence, however in adulthood they felt less stigma and improved self-perception, which was partially attributed to satisfactory surgical outcomes (Alansari et al., 2014). Research has additionally highlighted how some people recognise and report positive gains from living with and being treated for CL/P. Particularly mentioned is increased strength and maturity from their treatment experiences (Alansari et al., 2014), as well as improved social skills, sensitivity towards others (Eiserman, 2001) and increased social confidence (Stock et al., 2016).

In terms of overall adjustment to CL/P and associated challenges it seems the majority of those affected are able to adjust and cope well, with individuals reporting a limited impact of CL/P on friendships, romantic relationships or educational attainment and employment (Stock, Feragen, & Rumsey, 2015). Although a proportion of adults may describe ongoing distress and mental health difficulties which they attribute to CL/P related issues (Stock et al., 2015). The extent to which CL/P affects aspects of psychosocial wellbeing such as self-esteem, body image, quality of life and social interaction is variable (Rumsey & Harcourt, 2004) and not predicted by the severity of the visible difference (Rumsey, Clarke, & Musa, 2002). Rather, self-perception or investment in appearance and how this relates to self-worth seem to be better predictors (Crerand, Sarwer, Kazak, Clarke, & Rumsey, 2017; Alansari et al., 2014).

Reviews of the literature suggest evidence is somewhat inconclusive in determining whether CL/P patients are more at risk for psychosocial difficulties than unaffected peers (Hunt, Burden, Hepper, & Johnston, 2005; Stock & Feragen, 2016). The combination of known methodological challenges in the field and the lack of longitudinal studies contribute to the difficulty in being able to draw definite conclusions. This is in addition to the fact that psychosocial adjustment and functioning is a transient and multi-faceted concept and is thus hard to define and measure. The current consensus is that the effect of CL/P on an individual's quality of life and psychosocial adjustment appears to be generally low (Stock & Feragen, 2016), though there can also be extensive individual variation in adjustment to CL/P owing to underlying psychological and cognitive processes, and this may help to explain the inconclusive findings within the literature (Stock et al., 2016).

It is known that the CL/P specific psychosocial difficulties relate to dissatisfaction with cleft-related features and aspects of social functioning (Hunt et al., 2005; Stock & Feragen, 2016) which can subsequently effect wellbeing. A recent study found overall satisfaction with appearance to be similar across youth with and without CL/P, however those affected by CL/P reported more concerns about facial features (Crerand et al., 2017). As aforementioned, facial difference is seen as a vulnerability factor for stigmatisation or teasing (Hearst, Middleton, Owen, & Zeffertt, 2010; Strauss et al., 2007) and some youth indicate CL/P has affected their self-confidence (Turner et al., 1997). Discontent with facial appearance and desire for additional surgery has also been linked with worse mental health (Marcusson, Paulin, & Östrup, 2002; Sinko et al., 2005).

It is suggested that youth with visible differences may understandably be at increased risk of psychosocial issues due to the emphasis on appearance in this age group (Rumsey & Harcourt, 2007). However, there is also difficulty drawing firm conclusions about the impact of age on psychosocial adjustment, as many studies comprise wide age ranges and small sample sizes. A majority of studies have focused upon children and adolescents meaning relatively less is known about the psychosocial adjustment of young people as they progress into adulthood (Hunt et al., 2005). This transition period is important developmentally, as the significance of appearance, peer acceptance and relationships is heightened (Strauss et al., 2007). This is perhaps further amplified for those being treated for CL/P whose ‘cleft identity’ is highlighted by frequent appointments with the cleft team (Hearst et al., 2010). Narratives of adults treated for CL/P suggest that with age they became more accepting of residual CL/P features especially as other aspects of their lives took priority (Stock et al., 2016). Due to advances in treatment and surgery over time it is likely these adults may have had less options for additional ‘corrective’ surgery than patients today, which may or may not be a factor in their adjustment.

Elective CL/P treatment at the end of the pathway

It is during this transition between adolescence and adulthood that patients typically become more aware of, and make decisions about, the additional surgeries available to alter facial appearance or function and minimise cleft-related stigma (Cleft Lip and Palate Association, 2015; Goodacre & Swan, 2008). Unlike primary surgeries that are medically necessary and advised, secondary surgeries are elective and guided by a combination of clinical evidence and patient choice. Such surgeries are definitive in that they can dramatically alter appearance and primarily include revisions to the jaw, nose, and or lip, and are carried out toward the end of the 20-

year cleft treatment pathway once maturational and skeletal growth are anticipated to be complete. Undergoing appearance-altering surgery requires the integration of the revised facial features into one's identity and sense of self (Cash & Pruzinsky, 2002) and it has been recommended that the experience of surgery is investigated to explore the psychosocial impact (Hunt et al., 2005).

Considering the definitive nature of end of pathway cleft surgeries and given that they typically occur at a key transitional point in development, evaluating the impact of surgery upon patients is crucial in determining the value of interventions offered (Alansari et al., 2014; Eckstein, Wu, Akinbiyi, Silver, & Taub, 2011). Whilst clinician-reported outcomes can reveal part of the picture by considering treatment success based on clinical indicators, such outcomes are based on the clinician's perspective. Thus in recent years there has been growing recognition by researchers and clinicians of the value of capturing the patient perspective to evaluate outcomes (Aspinall, 2010; Nelson, 2009). Patient-reported outcomes (PROs) provide insights directly from the patient perspective about key health-related outcomes (e.g. health-related quality of life, psychosocial wellbeing, satisfaction, or function) of a particular treatment or intervention. The instruments which capture such outcomes can be generic and thus allow comparisons across groups or condition-specific and able to capture the pertinent issues relevant to that condition (Pusic, Lemaine, Klassen, Scott, & Cano, 2011). Accordingly, as historically less attention has been given to PROs the Systematic Review included herein aims to review the available literature on the PROs of end of pathway cleft surgery.

Decision making in paediatric healthcare

Ethically, it seems imperative that children are involved in decisions about matters that directly affect them, such as CL/P treatment and surgery (Bemmels et al., 2013). Four levels to decision making are suggested: 1) to be informed; 2) to express informed views; 3) to have views taken into account; 4) to be the lead decision-maker, if competent to do so (Alderson & Montgomery, 1996). This supports the idea that across the course of development children can be involved to differing degrees depending upon their level of competence. Therefore, it is anticipated that as children progress through the cleft pathway, and as their competence to make informed decisions matures, they will become more actively involved in treatment decisions.

Generally, however there is a lack of universal agreement about the age at which a child can be considered a competent decision-maker (Grootens-Wiegers, Hein, van den Broek, & de Vries, 2017). In some ways this is reassuring because decision making is dependent upon multiple contextual factors. Competence is not only specific to the decision in question but the situation and context, meaning a person competent to make a decision in one context may not be in another. For example, factors such as the emotional salience of a decision or situation is likely to impact on decision making. From a legal perspective, consent to medical treatment requires that the person is competent, that the decision is voluntary and that it is informed¹. It is suggested that to be deemed ‘competent’ the decision-maker should achieve the four standards of capacity, namely 1) to express a choice; 2) to understand the information; 3) to reason or weigh up the options presented; 4) to

¹ A decision can be defined as ‘informed’ when the decision-maker is aware of and uses all information about the pros and cons of all available treatment options in the context of their beliefs and what matters to them (Bekker et al., 1999).

appreciate the personal relevance of the available options (Appelbaum & Grisso, 2001; Grisso, Appelbaum, & Hill-Fotouhi, 1997).

It is proposed that the key factors influencing competence in making decisions are the child (in terms of their motivations, personality and interest), parents and clinicians (in terms of their attitude towards the decision) and the situation (in terms of the nature of the decision and time pressure), in addition to any predisposing factors for example, prior experience or cognitive development (Miller, Drotar, & Kodish, 2004). These factors are important when considering adolescent competence to make decisions as it suggested their competence may be more situationally variable and potentially more susceptible to the effects of emotion and peer influence (Grootens-Wiegers et al., 2017).

Shared decision making

One model or way of communicating, that values patient involvement in addition to clinical expertise, is shared decision making (SDM). SDM involves the clinician and patient working in partnership to mutually share knowledge, information and expertise, including discussion about the risks and benefits of available treatment options and the desired outcomes, in order to reach a shared decision (Charles, Gafnv, & Whelan, 1997; Coulter & Collins, 2011). Current healthcare policies in the UK advocate the use of SDM in routine clinical practice. For example, the National Institute for Health and Care Excellence (NICE) leads the Shared Decision Making Collaborative (a collection of organisations dedicated to embedding SDM in the healthcare system) and the NHS Right Care SDM programme is supporting the local and national implementation of SDM. It is recommended that all patients, including children, are involved in decisions about

their healthcare to respect the autonomy of patients by ensuring there is “no decision about me without me” (Coulter & Collins, 2011; Department of Health, 2012). As one characteristic of SDM is the focus upon establishing the patients values and preferences SDM can be especially useful in situations where the decision is weighted more towards personal or moral values than medical need.

In terms of making decisions about elective surgery a systematic review found SDM improved the quality of decisions made and it was also speculated that SDM may lead to more patients electing to not have surgery (Boss et al., 2016). It is suggested to facilitate SDM clinicians need to acknowledge the expertise, values and preferences of their patients by asking “what matters to you?” (Barry, Edgman-Levitan, & Billingham, 2012). A thematic synthesis on barriers and facilitators to shared-decision making (SDM) suggested patients need more than just knowledge of their personal values and the treatment. That is, patients need power (the awareness they can influence the decision making process) in addition to knowledge to be able to participate in SDM (Joseph-Williams, Elwyn, & Edwards, 2014). For YP in particular, who may be developing their autonomy, the ‘knowledge is not power’ paradigm may have particular relevance.

Theories of decision making

As humans we are required to make decisions all the time so the science of decision making has a long history. Various models of decision making exist including normative, prescriptive and descriptive models (Baron, 2012). Some models are more helpful than others in understanding how patients approach treatment decisions in a healthcare context, and in considering how we can facilitate decision making. For example, normative models such as those based on expected

utility have limited real-world applicability as they focus upon how ‘good decisions’ are made, and therefore assume the rationality of decision makers operating in idealised and unbiased contexts. Prescriptive theories focus on how individuals ‘should’ think in order to make informed decisions, and this focus on improving decisions has led to the development of decision aids, that are now widely used to support patients to make better decisions. Decision aids are designed to assist both patients and professionals in the decision making process by encouraging active participation and the presentation of unbiased, balanced information thereby allowing patients to assimilate information in line with their own preferences and values (Bekker et al., 1999; BMJ, 2013; O’Connor et al., 2003)

Perhaps the most useful and applicable to a healthcare context are descriptive theories of decision making which describe how we make real-world decisions based on our ability and motivation to process information. We tend to either process information systematically, which involves the conscious and deliberate processing and evaluation of all available information or heuristically, where practical short cuts are used to comprehend information (Chaiken, 1980). Due to limited cognitive capacity we tend to engage in heuristic modes of processing which enable us to simplify information, often by selectively attending to contextual aspects of the information presented (Chaiken, 1980; Marewski, Gaissmaier, & Gigerenzer, 2010; Simon, 1956). For instance, in a medical context we might employ a social heuristic and pay more attention to our beliefs about the person delivering the information, than we do to the information itself (Marewski et al., 2010). It is suggested that limited cognitive capacity and a variety of contextual factors mean patients tend to make treatment decisions using heuristic processing (Bekker, 2009; Marewski & Gigerenzer, 2012).

With this in mind and given that young people being treated for CL/P are required, at the end of the pathway, to make decisions about whether to undergo appearance altering surgery (e.g. Orthognathic Surgery) the empirical study presented herein aims to develop our understanding of their decision making experiences. This is especially pertinent for three reasons: namely the experiences of young people being treated for CL/P at the end of the pathway are under-represented in the field (Hall, Gibson, James, & Rodd, 2012; Hunt et al., 2005), secondly young people are making decisions about surgery that is likely to dramatically alter their appearance and thirdly the decision is being made at a developmentally sensitive time in their life.

Whilst classic models of decision making are important and can provide a theoretical basis to understanding the experiences of young people making decisions about orthognathic surgery, they seem unable to capture the complexities involved in making elective decisions about appearance altering surgery during a key transitional period of development. Other than explaining the likelihood of YP engaging in heuristic modes of processing they cannot account for the contextual, social and individual factors likely to affect YP's experience of decision making. Hence the empirical study adopted a qualitative approach to provide the necessary insight into what decision making is like for this population.

Chapter Two: Systematic Review

The patient-reported outcomes of end of pathway
cleft surgery: A systematic review.

Abstract word count: 232

Paper word count: 6991

Prepared in accordance with the requirements for submission to
The Cleft Palate-Craniofacial Journal (see guidelines in Appendix 1).

Abstract

Objective: To identify and review the literature on the patient-reported outcomes (PROs) of surgery at the end of the cleft treatment pathway.

Design: A systematic literature search was performed using electronic databases (Medline, PubMed, EMBASE, PsycInfo, Web of Science and Science Direct) from database inception to September 2017, to identify studies which measure and report the PROs of end of pathway cleft surgery.

Results: Of 263 identified papers 22 studies were deemed eligible for inclusion. Apart from one randomized controlled study, primarily studies were observational and adopted a cross-sectional or retrospective design. The methodological quality was variable with the majority (n= 16) being small scale studies and only one third (n= 8) achieving a 'good' quality rating. None of the included studies utilised a measure validated in the cleft population, with a high proportion of studies utilising bespoke measures. Although findings are tentative, the generally high levels of patient satisfaction reported suggest patients derive benefit from undergoing end of pathway cleft surgery.

Conclusions: Due to the methodological challenges and the heterogeneity of what, how and when outcomes are measured and reported it is difficult to determine the PROs of end of pathway cleft surgeries. Consequently, this review advocates the conduct of well-designed, longitudinal studies using cleft-sensitive tool/s to capture PROs of end of pathway cleft surgery at various time points.

Key words: cleft, surgery, patient-reported outcomes, satisfaction, appearance, rhinoplasty, osteotomy, review.

Introduction

From birth to young adulthood the primary and ongoing objective of cleft treatment and surgery is to normalise appearance and function to improve quality of life (Colbert et al., 2015; Marsh, 1990; NHS England, 2013; Wehby and Cassell, 2010). For many patients, surgery at the end of the cleft treatment pathway may be recommended or requested to optimise facial appearance and minimise stigma associated with cleft-related features (Cohen et al., 2009; Hearst et al., 2010; Marsh, 1990; Tiemens et al., 2013). As well as intensive orthodontic work, various secondary surgeries may be offered during this period of development (Cleft Lip and Palate Association, 2015; Goodacre and Swan, 2008). These include surgery to improve jaw alignment, (orthognathic surgery or distraction osteogenesis) and/or surgery to improve the appearance and function of the nose (rhinoplasty). Surgery can also be offered to optimise the appearance of the repaired cleft lip (lip revision) in addition to secondary speech surgery to minimise hyper-nasal speech.

Due to their definitive nature and effect on facial aesthetics, these surgeries typically take place at end of the treatment pathway, in the teenage and early adulthood years when structural growth of the face is considered complete (Cleft Lip and Palate Association, 2015; Kaufman et al., 2012; Robin et al., 2006; Stal and Hollier, 2002; van der Heijden et al., 2008; Wolford and Stevao, 2002). However, the heterogeneity within the published literature, underreporting and lack of longitudinal data means it is difficult to estimate the proportion of patients who undergo these surgeries (Sitzman et al., 2016).

To determine whether cleft treatment meets its primary objective, cleft outcome studies tend to favour measuring and describing the clinician-reported

outcomes, such as clinical indicators of surgical proficiency or aesthetics based on the professional perspective (Eckstein et al., 2011; Russell et al., 2011; Semb et al., 2005). Of course, measuring clinician-reported outcomes is important to establish the clinical effectiveness of treatment and ensure good practice, however the emphasis on clinician-reported outcomes means much less is known about the patient-reported outcomes (PROs) of end of pathway cleft surgeries (Hens et al., 2011; Impieri et al., 2017; Stock et al., 2015). It is acknowledged that PROs are especially important in determining the value of any healthcare intervention (Hens et al., 2011; Porter, 2010). Neglecting the patient perspective may put services at risk of providing sub-optimal care that may not meet patient's needs or expectations (Alansari et al., 2014; Aspinall, 2010; Nelson, 2009), not to mention the untold economical and societal consequences (Wehby and Cassell, 2010). In recent years there has been greater recognition of the need to consider the patient perspective in determining the true outcome of end of pathway cleft surgeries (Alansari et al., 2014; Aspinall, 2010; Eckstein et al., 2011; Ricketts et al., 2016; Semb et al., 2005)

Not only do end of pathway surgeries alter facial appearance or speech, they may affect an individual's sense of self, quality of life and social relationships. Undoubtedly, this complexity cannot be adequately captured by clinician-reported outcomes alone (Eckstein et al., 2011). This is further emphasised by studies which indicate considerable discrepancy between patient and clinician perception of treatment outcomes (Brattström et al., 2005; Meyer-Marcotty and Stellzig-Eisenhauer, 2009; Semb et al., 2005; Sinko et al., 2005). Moreover, prioritising PROs in this clinical population seems more pertinent when one considers end of pathway cleft surgeries are routinely discussed, and take place, at a pivotal point in the developmental trajectory. That is, a point where young people (YP) are forming

their identity and sense of self (Erikson, 1963), as well as prioritising social relationships and becoming more autonomous and independent (Hearst et al., 2010).

Rationale for present systematic review

To the author's knowledge no other published or ongoing systematic review exists on the PROs of end of pathway cleft surgery. This was confirmed by checking both PROSPERO and the Cochrane Database of Systematic Reviews in July 2017. Similar reviews focus upon psychosocial outcomes after Orthognathic Surgery (Hunt et al., 2001; Liddle et al., 2015; Soh and Narayanan, 2013) however exclude the cleft population due to their potentially differing expectations and perceptions to those with dento-facial deformity. The present review aimed to appraise the published studies reporting the PROs of patients who undergo end of pathway cleft surgery.

The present review

In synthesising the literature this systematic review aims to answer the following questions:

1. What are the PROs following end of pathway cleft surgery?
2. Does end of pathway cleft surgery result in benefits for patients?
3. Are the benefits transitory or long term?
4. How are PROs measured?
5. When are PROs measured?

Definitions. For the purposes of this review end of pathway cleft surgeries are defined as elective surgeries that typically occur in late adolescence and early adulthood, towards the end of the 20-year cleft treatment pathway (Cleft Lip and

Palate Association, 2015; Marsh, 1990), when growth is anticipated as complete². Such surgeries include Orthognathic Surgery (OS) and Distraction Osteogenesis (DO), rhinoplasty, lip revision surgery and secondary speech surgery (e.g. pharyngoplasty).

PROs are defined by use of measures that provide the patient perspective on key health-related outcomes of end of pathway cleft surgery such as measures of quality of life, psychosocial wellbeing and satisfaction. In the cleft population, satisfaction with appearance (or lack of) has been shown to relate to various aspects of psychosocial wellbeing (Hunt et al., 2005), including quality of life (Broder et al., 2017; Oosterkamp et al., 2007; Sinko et al., 2005), social functioning (Gkantidis et al., 2015; van den Elzen et al., 2012) and psychological resilience (Feragen et al., 2009). As such, clinicians and researchers often use satisfaction with appearance and surgical outcome as a proxy measure.

Method

Protocol development

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines (PRISMA; Liberati et al., 2009) were used to document the review process and results. The protocol was registered on the international prospective register of systematic reviews (PROSPERO; 2017 CRD42017071916).

Search strategy

Studies were identified using online databases, Medline, PubMed, EMBASE, PsycInfo, Web of Science and Science Direct. All available years were included,

² As the authors are psychologists and not from a medical background, guidance was sought from the cleft surgical team in order to define end of pathway cleft surgeries.

from database inception to September 2017. Guidance was sought from an experienced health librarian before performing the searches. Key search terms included ‘cleft*’, ‘orthognathic surgery’, ‘distraction osteogenesis’, ‘rhinoplasty’, ‘pharyngoplasty’, and ‘lip revision’ (and related surgical terms), in addition to terms related to PROs (e.g. ‘wellbeing’, ‘satisfaction’ and ‘quality of life’). Boolean operators (OR and AND) were used to expand the search and combine search terms. Specific craniofacial journals were also hand-searched for eligible articles, as were the reference lists of all included studies. Titles and abstracts of all identified articles were screened for inclusion using predefined criteria.

Eligibility criteria

Eligibility criteria was developed in line with PICOS Centre for Reviews and Dissemination guidance (CRD; Centre for Reviews and Dissemination, 2008) and was intentionally inclusive due to the limited research on this topic.

Inclusion criteria. All study designs were included where they included the PROs of cleft patients (both non-syndromic and syndromic) who had undergone end of pathway cleft surgery. All methods of outcome measurement were included, provided there was at least a post-surgery measure including satisfaction. Such broad inclusion criteria were set to enable evaluation of all methods for measuring PROs, to establish current practice and to determine the quality of available evidence (Stock and Feragen, 2016).

Exclusion criteria. Studies which provided PROs of only non-cleft populations were excluded. Where cleft patients were included as a subsample, studies were excluded when results were not filtered, due to inability to determine specific outcomes for the cleft sample. Studies which only included clinician or

observer-reported outcomes were excluded, as were studies which reported secondary cleft surgery undertaken before the end of the treatment pathway (or where it was unclear at what age the surgery was undertaken). Due to translation limitations, only studies published in English or with an English translation were included.

Study selection and data extraction

A first screening was performed based on title and abstracts. Full texts were retrieved for the second screening. To determine agreement for inclusion in the synthesis, full-text articles were reviewed according to the inclusion and exclusion criteria by two authors (MA, JY). Any discrepancies were discussed against the eligibility criteria to reach consensus. Data were extracted from included studies using a piloted proforma (Appendix 2) developed to capture pertinent study characteristics relating to the patient perspective. Where included, clinician-reported outcomes were not extracted due to this review aiming to determine the PROs of end of pathway cleft surgery. Key variables of interest included study design, sample size, participant characteristics (age and gender), type of surgery, method of measuring PROs, timing of outcome assessment and the PROs.

Quality assessment

Included articles were assessed for their methodological quality using an adapted version of the SIGN-50 rating tool (Scottish Intercollegiate Guidelines Network, 2011) which was developed in line with CRD guidance (Kmet et al., 2004) and tailored for the purposes of this review (Appendix 3). A second reviewer verified the quality ratings on a minimum 20 per cent sample of the papers (Appendix 4). Disagreements were resolved by referring to the quality criteria.

Results

Outcome of search process

The search identified 470 records and after removal of duplicates 263 records of interest remained. Screening of titles and abstracts led to the exclusion of 230 papers. Where it was not possible to determine relevance from the title and abstract alone, papers were accessed and screened in more detail (n=52). Full-texts were retrieved for 33 studies. Of these 22 were deemed eligible for inclusion by two authors (MA, JY). No additional studies were found from searching the reference lists of included articles. Following discussion and subtle refinement of the eligibility criteria to clarify matters of interpretation, agreement for inclusion was high (96%). Agreement was reached on the final paper by referring to the eligibility criteria. A total of 11 studies were excluded due to the absence of PROs, no surgery undertaken or not an end of pathway surgery or results not filtered for cleft patients (see Appendix 5). Figure 2 depicts the search strategy using a PRISMA diagram.

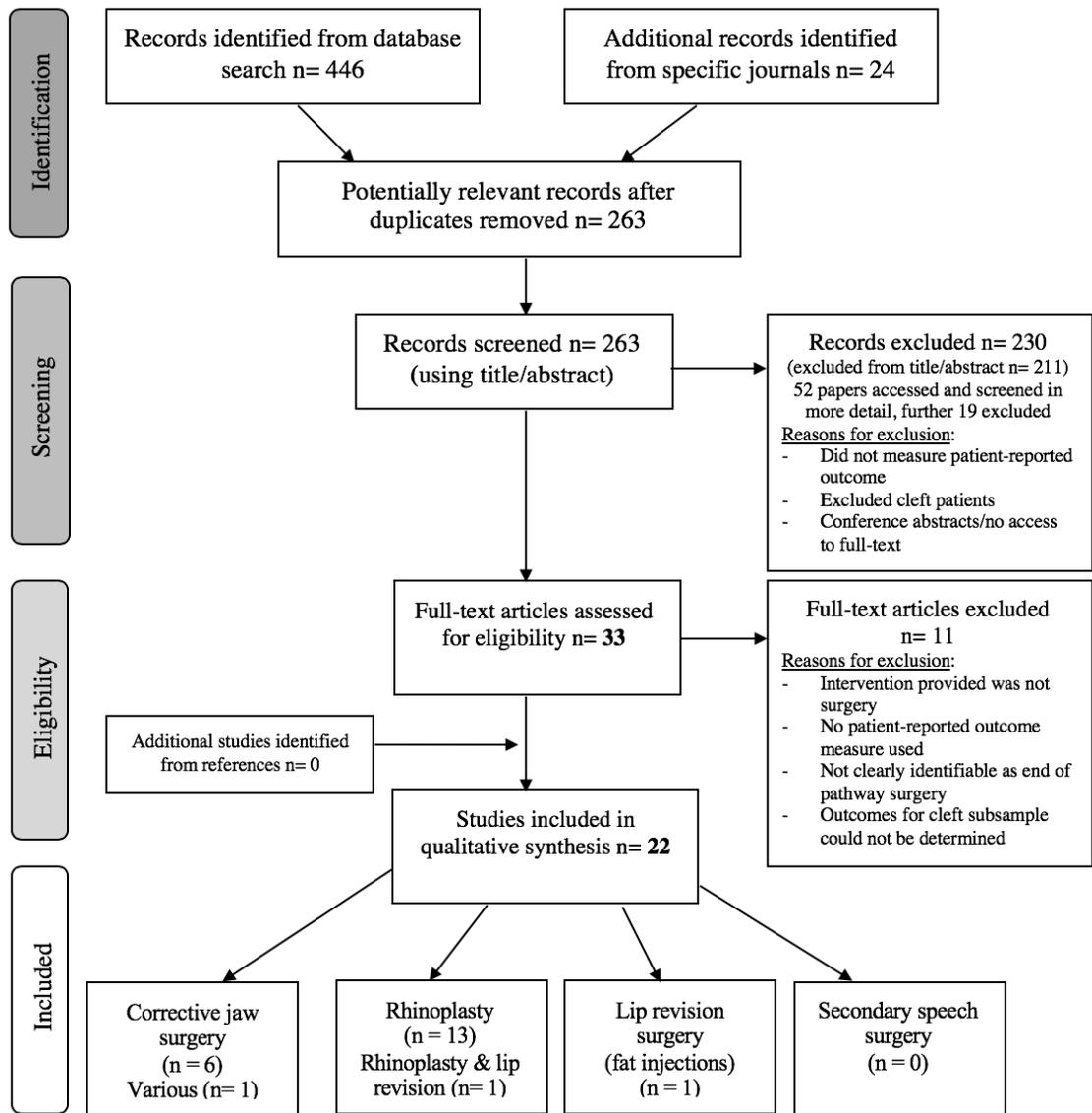


Figure 2. Outcome of search process

Study characteristics

Included studies were conducted in various countries across the world and all were single centre. The majority adopted an observational design (n=20). Of these, six were prospective studies (three with a comparator group), seven cross-sectional studies, and seven retrospective cohort studies (one with a comparator group). The review also found one randomised controlled study and one case study. This information is summarised in Table 1.

Over half the studies (n=13) measured PROs following rhinoplasty. Six studies measured outcomes of jaw alignment surgery (OS or DO). Two studies considered outcome following lip revision, one including rhinoplasty. One study considered outcomes following simultaneous end of pathway cleft surgeries. No studies measuring PROs following secondary speech surgery were eligible for inclusion.

The sample size varied (range, n=1-242) across studies, however most (n=16) were small scale with a sample size of less than 30 participants. Studies included both males and females, though tended not to analyse outcomes by gender. The mean age of participants across studies ranged from 16 to 32 years. Description of the surgical technique/s used was generally good, with only five studies not describing surgery in enough detail to permit replication.

Table 1. Level of evidence for PROs by study design					
Study design	Number of studies	Study	Sample size	Validated measure used? (Yes/No)	Number of times PROs measured
<i>Experimental</i>					
Randomised controlled	1	Chua et al. (2012)	30 (15 each surgery group)	Yes	3+
Non-randomised controlled	0	-	-	-	-
<i>Observational</i>					
Prospective cohort <u>with</u> comparator group	3	Cheung et al. (2006)	18 (9 cleft)	Yes	3
		Ricketts et al. (2016)	20	Yes	2
		Sawyer et al. (2017)	56 (27 cleft)	Yes	2
Prospective cohort <u>without</u> comparator group	3	Karabekmez et al. (2015)	9	No	1
		Roosenboom et al. (2014)	33	Yes	2
		Chaithanyaa et al. (2011)	10	No	1

Retrospective cohort <u>with</u> comparator group	1	Albers et al. (2016)	26	Yes	1
Retrospective cohort <u>without</u> comparator group	6	Eggermont et al. (2007)	9	No	1
		Vass et al. (2016)	12	No (modified)	1
		Hens et al. (2011)	30	Yes	1
		Sandor et al. (2006)	35	No	1
		Balaji et al. (2016)	21	Yes	1
		Jones et al. (2017)	11	No	1
Cross-sectional	7	Pausch et al. (2016)	242	No	1
		Gassling et al. (2015)	10	Yes	1
		Anderson et al. (2012)	19	No	1
		Byrne et al. (2014)	35	Yes	1
		Pitak-Arnop et al. (2011)	50	No	1
		Tiong et al. (2014)	16	No	1
		Scopelliti et al. (2013)	25	No	1
Case study	1	Simon et al. (2016)	1	N	1

Quality assessment of PROs

All included studies were rated for methodological quality, and a second rater independently reviewed the quality of five out of 22 studies. Exact agreement was initially achieved on 80% of criteria ratings and differed by one point on 20% (9 items). Differences were discussed, and total agreement reached by referring to, and more explicitly defining, the quality criteria to resolve matters of interpretation. As each criterion was not equally weighted the assigned quality scores do not represent a valid interval scale so are not reported herein due to the possibility of misinterpretation³ (Liberati et al., 2009). Instead studies are qualitatively categorised in line with SIGN-50 guidance based on the criteria fulfilled. A ‘Good’ rating

³ See Appendix 4 for the quality criteria rating table with inter-rater checks

indicates most of the quality criteria were fulfilled in relation to reporting of PROs; overall eight studies achieved this rating. Table 2 presents the summary of quality ratings.

Table 2. Overall quality ratings for included studies based on PROs

Quality category rating	Criteria	Studies
++Good	All or most quality criteria fulfilled, and if not, then conclusions unlikely to alter.	Chua et al. (2012). Ricketts et al. (2016). Pausch et al. (2016). Sawyer et al. (2017). Byrne et al. (2014). Roosenboom et al. (2014). Hens et al. (2011). Pitak-Arnop et al. (2011).
+ Acceptable	Some criteria fulfilled but some only partially fulfilled, or some criteria not met at all. Limitations may modestly affect the findings and conclusions.	Karabekmez et al. (2015). Andersen et al. (2012). Cheung et al. (2006). Eggermont et al. (2007). Albers et al. (2016). Vass et al. (2016). Gassling et al. (2015). Tiong et al. (2014). Chaithanyaa et al. (2011). Sandor et al. (2006). Balaji (2016). Jones et al. (2017).
- Poor	Few criteria fulfilled, concerns about methodological quality and ability to draw conclusions about PROs.	Scopelliti et al. (2013). Simon et al. (2016).

How were PROs measured?

Validated measures. Table 3 summarises the measures used to capture PROs. Most importantly, none of the studies included in this review used measures validated in the cleft population. It is however acknowledged that the availability of patient-reported quality of life or satisfaction measures that are validated in the cleft population is very limited (Eckstein et al., 2011). Only 40% of the studies utilised one or more validated patient-reported outcome measures. Three studies used multiple validated tools, likely in an attempt to capture a more comprehensive

understanding of outcomes from the patient perspective (Cheung et al., 2006; Chua et al., 2012; Roosenboom et al., 2014).

Table 3. Patient-reported outcome measures used

Measure used	Number of studies
Rhinoplasty Outcome Evaluation (ROE)	7 (1 modified)
Derriford Appearance Scale-59 (DAS-59)	3
Sheehan Disability Scale	1
Social Avoidance & Distress Scale (SADS)	2
Culture-Free Self-Esteem Inventory (CFSEI)	2
Satisfaction with Life Scale (SWLS)	2
Bespoke satisfaction questionnaire devised by author/s	15

Rhinoplasty Outcome Evaluation (ROE). The Rhinoplasty Outcome Evaluation questionnaire is a 6-item tool developed to evaluate patients' opinion on their pre and post-surgical nasal appearance and function across physical, emotional and social domains of life (Alsarraaf et al., 2001). It was developed to determine outcomes following cosmetic or post-traumatic rhinoplasty so has good test-retest reliability, internal consistency and responsiveness. Each item is scored on a 0-4 scale and the total score divided by 24 and multiplied by 100 to give a satisfaction score out of 100. Higher scores indicate greater satisfaction and a score of >85 indicates the patient is 'very satisfied'.

Derriford Appearance Scale-59 (DAS-59). The Derriford Appearance Scale is a validated questionnaire comprising 59 items and measuring the level of psychological distress or dysfunction related to physical appearance (Harris and Carr, 2001). It is a self-report measure designed for the adult population (≥ 16 years).

An introductory section asks respondents to identify the aspect of appearance of greatest concern. The DAS-59 derives a total score and five domain scores measuring aspects of self-consciousness; higher scores indicate greater distress about appearance. Standardisation tables allow patients' scores to be compared with clinical samples and with the normative population of those concerned and unconcerned about their appearance, and as discriminated by gender and age.

Social Avoidance and Distress Scale (SADS). The Social Avoidance and Distress Scale is a 28-item true-false self-report questionnaire which measures social anxiety, distress and avoidance behavior (Watson and Friend, 1969). Higher scores indicate increased social anxiety and distress.

Cultural-Free Self-Esteem Inventory (CFSEI). The Cultural-Free Self-Esteem Inventory (Battle, 1992) is a 60-item true-false questionnaire designed to measure an individual's self-esteem across four domains general, social, academic and parental. Higher scores suggest better self-esteem.

Satisfaction with Life Scale (SWLS). The Satisfaction with Life Scale is a 5-item questionnaire measuring general satisfaction with life using a 7-point Likert scale (Diener et al., 1985). Higher scores suggest greater life satisfaction.

Sheehan Disability Scale. The Sheehan Disability Scale (Sheehan, 1983) is a validated 3-item questionnaire that explores the generic quality of life in professional, social, and home life using a 10-point Likert scale. Higher scores are indicative of lower quality of life.

Bespoke questionnaires. Over half of the studies reviewed did not use a validated outcome measure and instead created a bespoke questionnaire. A number

of studies (n= 4) supplemented use of validated measure with an ad-hoc questionnaire; this may have been to counteract the shortcomings of generic measures and to ask questions of relevance to the cleft population (Eckstein et al., 2011). Questionnaires tended to ask questions about overall satisfaction with treatment outcome, satisfaction with facial aesthetic, effect of surgery on life, career, social world and interactions with others, willingness to undergo surgery again, and whether they would recommend the surgery to a friend. Some questionnaires asked patients to rate the facial feature that bothered them most before surgery and the feature most improved after surgery; this was most common in studies evaluating rhinoplasty.

Bespoke questionnaires differed in quality and in terms of the relative emphasis placed on functional and psychosocial outcomes of surgery. Attempts were made to generate relevant items, with one study interviewing three cleft patients who had undergone DO to gain insight into their experiences (Eggermont et al., 2007). Another reviewed the existing literature to support questionnaire development (Andersen et al., 2012). Whilst ad-hoc questionnaires can ask relevant questions, their reliability, validity and specificity cannot be determined (Eckstein et al., 2011). A number of authors recognised the limitations of using bespoke questionnaires but were motivated to measure patient perceptions as few prior studies had achieved this.

What are the PROs, are there benefits, and are these maintained?

Determining and summarising the PROs of end of pathway cleft surgeries is difficult due to the heterogeneity of what, how and when outcomes are measured and reported. Key outcomes are described below, and Table 4 presents a summary of findings from the included studies.

Table 4. Study characteristics and summary of key findings from included studies

Type of surgery	Study <i>Country of origin</i>	Sample size	Age (years) mean (\pm SD)	Gender	Patient-reported outcome measure/s	Timing of outcome assessment (Mean; range)	PROs
Orthognathic surgery	Karabekmez et al. (2015). <i>USA</i>	9/15 completed outcome measure	18.0 \pm 4.4	Not reported for those that completed questionnaire	Bespoke 20-item patient satisfaction with treatment and functional outcome questionnaire.	Post-surgery only (not reported)	Mean overall satisfaction level= 9.6 \pm 0.7 (where, 10= most satisfied). All 9 patients reported the surgery had a positive influence in their life or career and said they would have the surgery again.
Orthognathic surgery and bone graft	Simon et al. (2016). <i>India</i>	1	17	Female	Bespoke 1-10 satisfaction scale	Post-surgery only (not reported)	The patient gave 10 points (on 1-10 scale) for the treatment outcome and remained satisfied at 5-year telephone review.
Orthognathic surgery / maxillary distraction osteogenesis	Chua et al. (2012). <i>Hong Kong</i>	30 (15 each group)	Mean age not reported, range =18-22+	OS = 8 males, 7 females DO = 9 males, 6 females	Social Avoidance and Distress Scale (SADS) Cultural-Free Self-Esteem Inventory (CFSEI) Satisfaction with Life Scale (SWLS)	Pre and post-surgery and follow-up (2-8 weeks, 3 months, 6 months, 1 year, 2 years)	Both OS and DO surgeries resulted in a decrease in social avoidance and distress levels 2 years after surgery. Patients in each group had similar levels of self-esteem, social avoidance, and distress levels 2 years postoperatively, however those treated with DO reported higher levels of satisfaction with their lives after 2 years.

<p>Orthognathic surgery / maxillary distraction osteogenesis</p>	<p>Andersen et al. (2012). <i>Denmark</i></p>	<p>19 DO = 13 OS = 6</p>	<p>DO= 17.5 (±2.3) OS= 17.8 (±2.6)</p>	<p>Not reported for those that completed questionnaire</p>	<p>Bespoke 13-item patient satisfaction questionnaire concerning aesthetics and function, using VAS (0 – 100)</p>	<p>Post only (OS - 24.4months; DO - 24.8 months)</p>	<p>In both DO and OS a high level of satisfaction with the facial aesthetic was reported at the end of treatment. Both groups were satisfied with appearance according to themselves and the perceptions of relatives and others reaction. Both groups were satisfied with general wellbeing and social activities. The DO group expressed less satisfaction with treatment duration.</p>
<p>Orthognathic surgery / maxillary distraction osteogenesis (5 cleft patients had DO, 4 had OS; control group all had OS)</p>	<p>Cheung et al. (2006). <i>Hong Kong</i> <i>Single center</i></p>	<p>18 (9 cleft and 9 non-cleft)</p>	<p>Cleft patients = 18 (±2.24) Non-cleft = 22.56 (±5.86)</p>	<p>Cleft – 4 males 5 females Control – 2 males 7 females</p>	<p>Social Avoidance and Distress Scale (SADS) Cultural-Free Self-Esteem Inventory (CFSEI) Satisfaction with Life Scale (SWLS)</p>	<p>Pre (immediately before surgery), post (3-weeks) and 12-week follow-up</p>	<p>No statistically significant changes were reported over time on any of the measures. However cleft patients who underwent DO reported higher levels of social avoidance and distress and lower self-esteem than the OS and non-cleft group and this was maintained at 12-week follow-up. The presence and inconvenience of the distractor device, and prospect of further surgery may explain the poorer psychosocial outcomes of DO group.</p>

Distraction osteogenesis (external device)	Eggermont et al. (2007). <i>Netherlands</i>	9	17.7 (±4.0)	4 males 5 females	Bespoke satisfaction questionnaire with 4-point scale to rate satisfaction with appearance and function in 6 areas.	Retrospective pre, during and post-surgery (18.7; 6-28 months)	6/9 reported dissatisfaction with appearance before surgery including negative social interaction. 7/9 dissatisfied during treatment, received negative remarks and daily living, sleep, eating, personal care and affection were difficult. After surgery, 8/9 satisfied with appearance and had received positive remarks from others family. 5/9 said would undergo procedure again.
Rhinoplasty	Albers et al. (2016). <i>Netherlands</i>	26	31	13 males 13 females	DAS-59	Retrospective pre and post (3.9; 2-6 years)	Group of patients with cleft showed statistically significant post-surgery improvement in all scores of DAS-59, indicating decreased distress with appearance. Largest reduction on the Facial Self-Consciousness subscale, no significant effect of surgery found on negative self-concept scale. No significant difference between post-surgery scores of cleft group and 'concerned' normative group.

Rhinoplasty	Ricketts et al. (2016). <i>Canada</i>	20	30	11 males 9 females	DAS-59	Pre and at least 6 months post-surgery (11 months; 6 - 42 months)	Cleft patient's pre-surgery total DAS scores (male mean = 92; female mean = 112) were higher than the normative groups (male mean = 29.3 to 85.5; female mean = 22.0 to 115.3). Post-surgery, total DAS scores indicated a significant reduction in appearance-related distress. No significant changes were observed for the Facial Self-Consciousness or Physical distress and dysfunction subscales.
Rhinoplasty	Vass et al. (2016). <i>Hungary</i>	12	21	4 males 8 females	Modified 4-item ROE	Retrospective pre and post (4-6 months)	ROE total and individual item scores improved significantly post-surgery. Patients were satisfied with improved appearance of the nose and the opinion of others improved.
Rhinoplasty	Pausch et al. (2016). <i>Germany</i>	242	22.1	145 males 97 females	Bespoke 2-item measure of satisfaction with post-operative nasal appearance and function, using 3-point ordinal rating scale (good/moderate/bad), rated by patient and professional.	Post only (>6 months)	Patients reported good function (82 %) and good aesthetics (74 %). Professionals assessed aesthetics as good for 157 patients (65 %). Analysis revealed significant differences between patient satisfaction and professional assessment ($\kappa = 0.385$; $P < 0.0001$)

Rhinoplasty	Sawyer et al. (2017). <i>United Kingdom</i>	27 cleft (29 non-cleft)	Cleft =22 (± 9.8) Non-cleft (29 ± 14.9)	11 males 11 females (non-cleft – 19 males, 8 females)	ROE	Pre and post-surgery (4 months; 3-6 months)	Statistically significant improvement in pre-post mean ROE scores in cleft group (pre 28 ± 10 ; post 80 ± 11 ; $p < .01$) indicating significant improvement in appearance of nose post-surgery. All patients reported they would undergo the surgery again.
Rhinoplasty	Gassling et al. (2015). <i>Germany</i>	10	21 (median)	6 males 4 females	ROE	Post only (not reported)	90% return rate. Patient satisfaction was high (median score =87.5%, where >85 = high satisfaction). Majority were satisfied with appearance of nose, felt others liked it, felt surgery positively affected social and professional activities and did not desire further surgery. Higher satisfaction found in females.
Rhinoplasty	Byrne et al. (2014). <i>Ireland</i>	35	27.6 \pm 11.7	16 males 19 females	ROE Bespoke Preoperative and Postoperative Semi-quantitative Ordinal Rating Scale of nasal appearance Bespoke 7-item semi-structured questionnaire about satisfaction with	Post-surgery (2.6 years, 16 months - 5 years)	Post-surgery patient satisfaction was high as measured by the ROE, (score 76.1). Teenage female's (<20 yrs) satisfaction was higher than older females or males. On the ordinal scale 91% rated their appearance as improved. 34/35 said

					nasal appearance and surgery.		would undergo surgery again.
Rhinoplasty	Roosenboom et al. (2014). <i>Belgium</i>	33	22.1	24 males 9 females	VAS for nasal function & appearance ROE DAS-59 Sheehan Disability Scale	Pre and post-surgery (1 year)	High patient satisfaction 12 months after rhinoplasty with significant improvement in quality of life. ROE score significant post-surgery improvement (p < 0.0001) DAS-59 - significant decrease of self-consciousness in all areas except facial self-consciousness (p = 0.14) Sheehan Disability Scale - total quality of life was significantly higher than before surgery (p = 0.01), especially at home and in social situations (p = 0.008 and p = 0.03, respectively) 94% said would undergo surgery again.
Rhinoplasty	Hens et al. (2011). <i>Belgium</i>	30	27.2	15 males 15 females	ROE Bespoke satisfaction with nasal appearance and function (1-10 scale)	Retrospective pre and post (6 months-3 years)	Significant post-rhinoplasty improvement in mean ROE score, from 39.3 ± 3.1 preoperatively to 73.1 ± 2.0 postoperatively. 29/30 rated their nasal appearance as better, 18/30 rated nasal function as improved. All patients

							would undergo the surgery again knowing the final result.
Rhinoplasty secondary cleft rhinoplasty (with versus without a caudal septal extension graft)	Pitak-Arnop et al. (2011) <i>Germany</i>	50	19.8 ± 5.3	26 males 24 females	Bespoke preoperative and postoperative semi-quantitative ordinal rating scale of nasal appearance. Bespoke 4-point satisfaction questionnaire (4=very satisfied).	Post only (not reported)	90% revealed their improved nasal aesthetics postoperatively and expressed satisfaction with 3 or 4 on scale. Patient's satisfaction did not correlate with patients age or gender, surgical technique, or panel perception of surgical outcome
Rhinoplasty	Tiong et al. (2014) <i>Malaysia</i>	16	20.4	5 males 11 females	Bespoke satisfaction questionnaire measuring patients' perception of their cleft lip nasal deformity before and after operation and overall satisfaction on 0-10 VAS.	Retrospective pre and post. (not reported)	Preoperatively 87.5% of patients reported nasal asymmetry as main complaint, and postoperatively 75% reported good or excellent improvement. On the VAS 93.7% scored overall satisfaction with surgery between 5-8 (where 10 = most satisfied). All patients would recommend the surgery.
Rhinoplasty 6/10 had undergone OS to improve profile prior to rhinoplasty	Chaithanyaa et al. (2011) <i>India</i>	10	20.4	3 males 7 females	Satisfaction questionnaire using VAS	Post-surgery only. 1 year.	Patients reported satisfaction with both their post-operative function and appearance. Patients were most satisfied at the nasal tip followed by the dorsum, unequal alar bases and

							nostril. All patients satisfied with the outcome of the surgery and said would undergo again.
Rhinoplasty (external)	Sandor et al. (2006) <i>Canada</i>	35	32.4	20 males 15 females	Satisfaction questionnaire and VAS (0-10) to rate pre and post-surgery appearance at 4 anatomical sites	Retrospective pre and post-surgery only. 28 months, 24–30 months)	Patients reported improved post-surgery appearance, with the highest VAS score at the nasal tip. All 35 patients would undergo surgery again.
Rhinoplasty & lip revision with abbe flap	Balaji (2016). <i>India</i>	21	22.87 ± 4.23	13 males 8 females	ROE Patients assessment of post-rhinoplasty success	Retrospective pre and post (6 months - 3 years)	Statistically significant change in mean ROE score (P = 0.001) from pre-surgery (19.8 ± 11.2), to post-surgery (78.5 ± 21.2). Post-rhinoplasty success – most patients perceived their post-surgery difference as unnoticeable, 9/21 perceived their scar as obvious to themselves but not others, though no patients desired further revision.
Lip revision with fat grafting (1/2 had simultaneous rhinoplasty)	Jones et al. (2017) <i>USA</i>	11/18 completed satisfaction measure	16.1	5 males 13 females	Bespoke 5-item satisfaction questionnaire with 5-point Likert scale	Retrospective pre and post (not reported)	Patients were positive about their experience and there was a significant post-surgery improvement in satisfaction with appearance (p< .001). All patients said would undergo surgery again.

Simultaneous Osteotomy with other revision surgeries	Scopelliti et al. (2013) <i>Italy</i>	25	>18 years	21 males 4 females	Bespoke satisfaction questionnaire	Post only (not reported)	Results unclear. 96% of patients satisfied with the jaw surgery and favorable for combined surgery. 88% satisfied with lip-nose surgery. 76% would advise to a friend.
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^aAbbreviations: OS= Orthognathic surgery; DO= Distraction Osteogenesis; ±SD= ±standard deviation; VAS= visual analogue scale; ROE= Rhinoplasty Outcome Evaluation; DAS-59= Derriford Appearance Scale-59.

Use of validated tools to measure satisfaction with life

Two studies prospectively measured patients' satisfaction with jaw re-alignment surgeries by using a validated but generic satisfaction with life tool (SWLS; Diener et al., 1985). One study did not find significantly increased levels of satisfaction at 12-weeks post-surgery for either type of jaw re-alignment surgery (Cheung et al., 2006). The other study found the DO group reported higher eventual levels of satisfaction than the OS group at 2-years post-surgery, however both groups were already 'slightly satisfied' before surgery (Chua et al., 2012). Timing of outcome measurement therefore appears important.

Use of validated tools to measure satisfaction with appearance

Seven of the 13 studies evaluating rhinoplasty outcomes measured satisfaction with nasal appearance and function by using the validated ROE questionnaire (Alsarraf et al., 2001). Results from the two prospective studies report significant pre to post-surgery improvements in ROE scores indicating greater satisfaction with nasal appearance after rhinoplasty, as measured at 4 or 12 months respectively (Roosenboom et al., 2014; Sawyer et al., 2017). Sawyer et al. (2017) found cleft patients' post-surgery scores were comparable to non-cleft rhinoplasty patients, which the authors suggest is promising given cleft rhinoplasty represents a greater surgical challenge. The other studies report similarly high post-surgical satisfaction with nasal appearance, though two were retrospective and asked patients to complete both the pre and post ROE after surgery (Balaji, 2016; Hens et al., 2011), which may have biased results in favour of surgery. Balaji (2016) did provide patients with pre-surgical profile photographs to aid

their memory in completing the pre-surgery measure, which may have unintentionally further biased results.

Two studies based their findings on a single post-surgery measure of satisfaction (Byrne et al., 2014; Gassling et al., 2015). The lack of a pre-surgery measure severely limits the conclusions that can be drawn from both studies. Furthermore, one study did not report how long after surgery outcomes were measured, meaning the validity of findings is questionable (Gassling et al., 2015). The authors acknowledge that the high rates of post-surgery satisfaction may be less related to the surgical outcome and more related to social desirability given the longstanding relationships patients establish with their cleft team. Indeed, patients' responses were not anonymous in Roosenboom et al. (2014) which may have positively biased results, although this is somewhat mitigated by it being a prospective study.

A further study also reported significant post-surgery improvements (Vass et al., 2016), however aside from it being a small, retrospective study with questionable use of inferential statistics, there are concerns about the validity of the findings due to modification of the ROE from four to six items. The rationale for modification is unclear and led to the exclusion of two items concerning confidence with appearance and desire to undergo surgery to alter the nose. Given the shortcomings of this study one may have expected the authors to be more cautious when drawing conclusions.

Measuring satisfaction using bespoke tools

Other measures of satisfaction were largely bespoke; therefore any results should be interpreted cautiously as the measures are likely to lack validity, reliability and

responsiveness to change over time. Generally speaking, results from bespoke satisfaction questionnaires indicate a high degree of patient satisfaction with end of pathway surgery. To measure satisfaction with jaw re-alignment surgery one study created a bespoke 20-item questionnaire to determine satisfaction with treatment and function (Karabekmez et al., 2015). The mean overall satisfaction was 9.6 ± 0.7 (where, 10= most satisfied). All nine patients confirmed a positive effect of surgery on their life or career and all said they would undergo surgery again; however, satisfaction was only measured post-surgery. Additionally, the questionnaire response rate was 60% and possible reasons for this are not discussed. Although most of the items related to functional outcomes, it is encouraging the authors included a question about the social impact of the surgery. In a similar vein, though using a visual analogue scale (VAS), Andersen et al. (2012) asked a number of questions pertaining to self and others satisfaction with aesthetic outcome, as well as the effect of surgery on wellbeing and social activities. Patients undergoing both types of jaw surgery (DO and OS) reported high levels of satisfaction across all areas, though the DO group were more dissatisfied with treatment duration. Dissatisfaction was observed in another DO study (Eggermont et al., 2007) which also included questions pertaining to the social impact. This study found 8 out of 9 patients were satisfied with their post-surgical appearance. The only case study included in this review reported high patient satisfaction (10/10, where 10 = most satisfied) with treatment (Simon et al., 2016), and suggest this was maintained at 5-year telephone review. However, the lack of detail about the measure used or when it was administered perhaps indicates prioritisation of surgical outcomes and makes it hard to interpret findings.

Of the studies using bespoke measures to assess rhinoplasty outcomes, the study in this review with the largest sample size (n=242) used a crude 2-item measure of post-surgery satisfaction with appearance and function (Pausch et al., 2016). On a 3-point scale (good/moderate/bad) 82% patients reported good function and 74% reported good aesthetic outcome. Unfortunately, as there was no pre-surgery measure we cannot determine whether this finding represents a significant change. However, it is acknowledged the study aimed to consider the relationship between patient and clinician-reported outcomes rather than assess pre to post surgery change. In addition, 71 patients declined to participate, and the authors considered that those who participated may have been more satisfied.

Some bespoke measures asked patients to rate their satisfaction at specific anatomical sites of the nose with the nasal tip and symmetry reported to be the areas of most concern pre-surgery and the most improved sites post-surgery (Chaithanyaa et al., 2011; Sandor and Ylikontiola, 2006; Tiong et al., 2014). Other questionnaires asked patients to rate their overall satisfaction with nasal appearance pre and or post-surgery and again, a high proportion of patients reported improved post-surgical appearance (Byrne et al., 2014; Chaithanyaa et al., 2011; Hens et al., 2011; Pitak-Arnop et al., 2011) however in many cases there is limited statistical evidence to confirm the significance of these findings.

The satisfaction ratings appear more variable in one study (Tiong et al., 2014) with overall satisfaction ratings ranging between 5-8 (where 10=most satisfied); this might be explained by the type of rhinoplasty undertaken and the fact there were some surgical complications. The study also does not make clear when the satisfaction

measure was administered which could have affected ratings. One retrospective study used a 3-point rating scale of post-rhinoplasty success (unnoticeable, obvious, deformed) and found most patients perceived their post-surgery visible difference as unnoticeable, with 9 out of 21 perceiving their scar as obvious to themselves but not others (Balaji, 2016).

It is also relevant to consider that in the Jones et al. (2017) study half the sample underwent a secondary rhinoplasty at the same time as lip revision, although the study was primarily measuring outcomes of fat grafting. Thus, it may be hard to ascertain whether outcomes pertain to the specific surgery being evaluated or the accumulated outcome of multiple surgeries at that point in time. One study did aim to measure outcomes following simultaneous surgeries, however the results are only briefly reported and are somewhat unclear in terms of what surgeries the results pertain to, as well as the response rate (Scopelliti et al., 2013).

To determine overall satisfaction, a number of studies asked patients whether they would be prepared to undergo surgery again knowing the outcome, and a high proportion of patients said they would, range 94-100% (Byrne et al., 2014; Chaithanyaa et al., 2011; Hens et al., 2011; Jones et al., 2017; Roosenboom et al., 2014; Sandor and Ylikontiola, 2006) or they would advise a friend to undergo the surgery (Scopelliti et al., 2013; Tiong et al., 2014).

Social distress

Two studies measured social distress using the SADS. Results from the randomised controlled study found that both types of jaw surgery resulted in similar and

decreased levels of social avoidance and distress at 2-year follow-up (Chua et al., 2012). No significant differences in SADS ratings were found between the two surgery groups. However, as the attrition rates for completion of the measure are not explicit it is difficult to ascertain whether results are based on a smaller subsample, thereby affecting validity. The other study measured outcomes before and at 3 and 12-weeks post-surgery and no significant post-surgery changes were observed for social distress (Cheung et al., 2006). However, the DO group reported consistently higher levels of social avoidance and distress compared to the OS and control groups, which is unsurprising given the presence of the externally visible distractor device⁴ and short duration of post-surgical assessment.

Appearance-related distress

Studies measuring this concept used the DAS-59 and infer that end of pathway cleft surgery reduces appearance-related distress. However, this conclusion is limited to three studies, and while two are prospective in design all studies only measure rhinoplasty outcomes and draw findings from small samples ($n \leq 33$). For example, one study found a significant post-surgery improvement in appearance distress as measured by DAS-59, and the post-surgery scores matched with those of the normative group 'concerned' about appearance (Albers et al., 2016). Most change was found on the facial self-consciousness subscale and no change was found on the negative self-concept subscale. However, this study is limited by the retrospective design and the variation as to when post-surgery outcomes were collected (2-6 years).

⁴ The aim of Distraction Osteogenesis is to make a bone longer by cutting the bone and using a device called a distractor to gradually pull apart the bone segments and generate new bone growth.

A different study found that pre-surgery cleft patients reported greater appearance-related distress than their non-cleft counterparts ‘concerned’ about their appearance. Outcomes measured at least six-months post-surgery revealed a significant reduction in overall appearance-related distress (Ricketts et al., 2016). This is in contrast to Albers et al. (2016) where no significant changes were observed for the facial self-consciousness or physical distress subscales. Similar findings were reported by Roosenboom et al. (2014) where appearance-related distress improved significantly post-rhinoplasty, across all areas apart from facial self-consciousness. Unfortunately, unlike in the Albers et al. (2016) study Ricketts et al. (2016) did not compare post-surgery scores with normative data, so it is difficult to determine the clinical significance of the change. However, conduct of an in-depth item-by-item analysis revealed that of the 59 items only 11 showed significant change and were likely to be measuring concepts relevant to the cleft population (Ricketts et al., 2016). Although limited by the small sample, this study should be credited for analysing change on individual items to consider the validity of the findings based on the measure used. Indeed, the DAS-59 is a generic tool and evidently lacks sensitivity to measure issues relevant to the cleft population.

Self-esteem

Only two studies measured self-esteem using a specific, validated measure, namely the CFSEI. Findings from these studies, which both prospectively measured self-esteem in relation to two types of jaw surgery appear inconclusive. The randomised controlled study found social self-esteem during the early post-surgery period improved for the OS group but reduced for the DO group (Chua et al., 2012). This difference is

likely attributable to the OS group noticing a more immediate surgical change. Whether there was a significant change in pre to post surgery self-esteem remains unclear, as it was only reported that both group's self-esteem reached similar levels 2-years post-surgery. The other study did not find a significant post-surgery change in self-esteem (Cheung et al., 2006), however the sample size was small and the duration of follow-up may have been too short to detect any change. Although this self-esteem tool is widely used in Hong Kong it has come under criticism for not being free of cultural bias (Brooke, 1995; Holaday et al., 1996).

Quality of Life (QoL)

Quality of life was overtly measured in one study (Roosenboom et al., 2014). Using the Sheehan Disability Scale this prospective study found general QoL was significantly higher at one-year post-surgery ($p= 0.01$), as was QoL at home ($p= 0.008$) and in social situations ($p= 0.03$).

Summary

Findings from the included studies suggest that patients derive some benefits from undergoing an end of pathway cleft surgery. This is most clearly demonstrated by high levels of reported patient satisfaction as well as some specific psychosocial outcomes related to improved quality of life, social interactions and decreased appearance-related distress. However, for the most part these conclusions are drawn from studies of questionable quality and methodological rigour, with a large number of retrospective and cross-sectional studies only measuring PROs at one point in time and using non-validated measures. Consequently, such conclusions need to be interpreted with caution.

Timing of outcome assessment and duration of follow-up

Studies differed in relation to the timing of outcome assessment and duration of follow-up. Five studies collected data prospectively (pre and post-surgery), and two studies also measured outcomes after one or more follow-up periods, ranging from 12-weeks to 2 years. The remainder of studies in this review either collected retrospective pre and post data or only collected post-surgery outcomes, thereby introducing an obvious potential for bias. There was much variation across the studies as to when post-surgery outcomes were measured, with the earliest reported measure being 3-weeks, and the longest 6-years post-surgery. The prospective cohort study measuring PROs at 3 and 12-weeks post-surgery did not find any significant post-surgery changes (Cheung et al., 2006). This is perhaps unsurprising given patients would still be healing physically as well as psychologically adjusting to their altered appearance. This study also administered the pre-surgery measures immediately before surgery which may have introduced a bias with patients amplifying distress or concern about appearance to justify undergoing surgery. Unfortunately, seven studies did not report the time period when post-surgery outcomes were collected (Karabekmez et al., 2015; Simon et al., 2016; Gassling et al., 2015; Pitak-Arnnop et al., 2011; Tiong et al., 2014; Jones et al., 2017; Scopelliti et al., 2013).

The inconsistency in timing of outcome assessment across the studies, poor reporting, lack of follow-up data and apparent biases makes it difficult to ascertain when benefits emerge, to compare PROs between studies and to ascertain whether outcomes are maintained or change over time. The optimum post-surgery window in which to measure outcomes (accounting for surgical healing, adjustment and minimising loss to

follow-up) is not clearly defined in the literature. This lack of guidance may mean that at worst, clinical teams fail to measure PROs or at best, measure them ineffectively.

Sawyer et al. (2017) recognised how their study could have been improved by measuring post-surgical outcomes later, but they were concerned about loss to follow-up. Another study recommended evaluating patient satisfaction preoperatively and at one and two-years post-surgery to allow adequate time for patients to adjust to changes and reflect on the effect of surgery (Sandor and Ylikontiola, 2006).

Discussion

This review of 22 studies aimed to appraise the literature on the PROs of end of pathway cleft surgery. The findings indicate there are relatively few studies which measure PROs, and the studies which do are relatively recent which is reflective of the changing culture of healthcare and growing recognition of the importance of the patient perspective. Findings from this review indicate that whilst patients do appear to derive benefit from the surgery, the heterogeneity between studies in terms of what, how and when outcomes are measured makes drawing valid conclusions about PROs difficult.

Over half of the studies considered PROs of rhinoplasty. This might be explained by the nose often being a particularly stigmatised feature and the one of most concern to patients (Marsh, 1990; Semb et al., 2005; Wong Riff et al., 2017) not to mention rhinoplasty being the most requested surgery (Sinko et al., 2005). In addition, the availability of a validated measure (ROE; Alsarraf et al., 2001) makes it easier to evaluate PROs (Sawyer et al., 2017).

Unfortunately, the ROE is not validated in a cleft population and accordingly none of the studies in this review used a patient-reported outcome tool validated in the cleft population. The subsequent use of generic measures likely led to an inability to capture the pertinent PROs and domains of life specific to the cleft population undergoing end of pathway surgery. This is evidenced by the item level analysis revealing few items of salience to a cleft population on a measure of appearance-related distress (DAS-59; Ricketts et al., 2016). A review of existing measures revealed just five QoL measures that are validated in the cleft population (Eckstein et al., 2011). However, only one of the five measures were specifically designed for the cleft population and involved patients in its development. Moreover, across all five identified measures there are content omissions in terms of surgical and treatment experiences and cleft specific functional difficulties (Eckstein et al., 2011). In the context of measuring PROs at the end of the pathway the Youth Quality of Life-Facial Differences questionnaire (Patrick et al., 2007) is the only measure which can be used with patients up to the age of 18 years as the other measures are designed either for parents or for patients aged 14 years and under.

Whilst it is recognised that the crude or generic measures used by the studies in this review were unable to adequately capture PROs of surgery (given the complexity of factors involved), considering there is no condition or procedure specific measure available, the studies in this review might be commended for attempting to measure PROs. Importantly though, measuring PROs was not the primary aim in a number of studies included in the review. Instead their objective may have been to report the surgical technique (Jones et al., 2017; Tiong et al., 2014), compare patient and

professional ratings (Pausch et al., 2016) or correlate patient satisfaction with other variables (Pitak-Arnop et al., 2011). Therefore, less consideration was given to how to effectively measure PROs, perhaps explaining the high proportion of studies utilising a non-validated measure. It also meant in some cases, even where a prospective design was adopted, the opportunity to measure PROs before surgery was missed (Chaithanyaa et al., 2011; Karabekmez et al., 2015). This observation reinforces how the field remains somewhat dominated by a focus upon clinician-reported outcomes. However even in the clinician-reported assessment of cleft outcomes there remains limited international consensus (Al-Omari et al., 2005) and the wide variety of measures available suggests the lack of a valid measure (Sharma et al., 2012). With their specialist training and knowledge about patient adjustment, QoL and psychosocial wellbeing psychologists are well placed to support clinical teams in evaluating end of pathway surgeries by advising on the use of appropriate tools to measure PROs.

Undoubtedly, as recognised by Eckstein et al. (2011) there is need for development and validation of a cleft-specific tool to measure PROs related to patient satisfaction, psychosocial wellbeing and health-related QoL. We would recommend that measures also be designed to capture end of pathway surgery outcomes. Currently under development and evaluation is the CLEFT-Q, an international patient-reported outcome tool designed to evaluate cleft outcomes across the age range (8-29 years) and across key domains of importance to the cleft population – appearance, health-related QoL and facial function (Tsangaris et al., 2017; Wong Riff et al., 2017). As each of the scales will be independent, the CLEFT-Q can be adapted for use in clinical and research settings. This tool shows promise in allowing the evaluation and understanding of the

patient perspective. It is determined to be valid, reliable and responsive the CLEFT-Q would benefit from routine use within and across services and research settings to inform clinical practice and measure quality of care. However, as aforementioned, there needs to be clarity on the time-points when outcomes should be measured in order that we are able to reliably and prospectively capture PROs, in addition to satisfaction with surgical outcome.

To the author's knowledge this is the first review in this area and due to the extant literature it is deliberately comprehensive and inclusive of the available literature, to both permit exploration of current understanding (Stock and Feragen, 2016) and to make recommendations for future research. A key observation in the present review, and one consistently noted in other related reviews (Hunt et al., 2005; Stock and Feragen, 2016), is the methodological flaws evident in the majority of studies. Thus, reliably evaluating PROs is challenging as studies tend to adopt retrospective or cross-sectional designs and utilise small samples. This is likely because retrospective and small-scale studies are easier and less costly to conduct, however the derived outcomes may be inaccurate and affected by recall bias (Song and Chung, 2010). The only randomised controlled study (Chua et al., 2012) in this review represents the highest level of evidence in terms of accepted definitions of methodological quality (CEBM, 2009). However randomized controlled trials are not well suited to surgical interventions due to ethical challenges (Chung and Burns, 2008). Song and Chung (2010) suggest well-designed observational studies are better suited to address questions in plastic and reconstructive surgery and can produce comparable results.

Strengths and limitations

This review considered two neglected areas in the field, namely PROs and cleft surgery at the end of the treatment pathway. Secondly, this review provides justification for the development of a cleft-specific tool and for the conduct of well-designed studies to more accurately determine the PROs outcomes of end of pathway surgery. The quality of this review was enhanced by two reviewers determining study eligibility for inclusion and by performing an inter-rater quality check. To increase validity a second researcher could have independently extracted data or checked the proforma for accuracy.

Due to time and resource constraints grey literature was not included so this review is limited by the effect of publication bias. It is also biased in favour of English language studies, however does include studies conducted across the world. Finally, contacting authors of primary studies to clarify or gather additional data may have been useful, especially because two studies were excluded on the basis that the cleft subsample data could not be determined.

Clinical implications and recommendations for future research

As psychology is now embedded within the cleft multidisciplinary team (Scott et al., 2014), psychologists can support medical colleagues in using appropriate measures to routinely evaluate PROs to help tailor care to patients' needs. Using cleft-sensitive tools to measure PROs related to surgery would also offer insight as to the value of surgical interventions.

An important finding was the variation as to when post-surgery outcomes are measured. Clinical consensus on the issue of when to measure outcomes and the recommended period of follow-up would be welcome to inform practice and guide research. To help with this, further research may wish to measure PROs at different time points post-surgery in order to map potential change and identify time-points where patients may be more or less likely to need psychological support or guidance.

As highlighted in other reviews (Hunt et al., 2005; Stock and Feragen, 2016) there is a need for high-quality prospective, longitudinal studies which measure the psychosocial status of cleft patients before, and for a significant period after end of pathway surgery. Ultimately, to establish whether end of pathway cleft surgery is beneficial to patients' psychosocial wellbeing, and whether changes are maintained, or indeed whether further surgery or intervention is sought, the field would benefit from conducting longitudinal studies. In order to maximise the contribution to the field, studies need to be well reported and researchers are encouraged to refer to the STROBE guidance (Vandenbroucke et al., 2007) when designing and reporting observational studies.

Conclusions

The present systematic review contributes to current knowledge and understanding by highlighting the ongoing difficulty in both determining and measuring PROs of end of pathway cleft surgeries. Although there is limited evidence suggesting patients derive benefit from end of pathway surgery the specifics remain unclear, and due to methodological challenges and a lack of evidence many claims are

unsubstantiated. A first step in determining the PROs of end of pathway cleft surgeries would be to focus effort upon *how* and *when* outcomes should be measured.

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Chapter Three: Empirical paper

On my shoulders: Young people's experience of the decision making process for orthognathic surgery in cleft lip and palate.

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The Cleft Palate-Craniofacial Journal (see guidelines in Appendix 1).

Abstract

Objective: To understand how young people (YP) being treated for cleft lip and or palate (CL/P) experienced the process of making a decision about whether or not to undergo orthognathic surgery (OS).

Design: The study adopted a qualitative design. Individual semi-structured interviews were conducted with YP who had made a decision about orthognathic surgery in the context of CL/P.

Participants/Setting: Twelve YP (mean age 21 years, 2 months) were recruited from two specialist cleft centres in the UK. Ten participants had decided to undergo OS and two decided not to. Interviews were conducted at participants' homes or local cleft centre.

Results: Thematic Analysis led to the development of four key themes: 1) *Awareness of difference*, 2) *Committing to the process* 3) *Others facilitating decision making* 4) *Responsibility on my shoulders*.

Conclusions: Findings offer insight into the largely unexplored experiences of YP being treated for CL/P. For many, deciding represented a milestone in terms of cleft treatment and being an adult. Their experiences reveal the contextual, social and personal complexities involved in the process of deciding about OS during a period of developmental transition. This study highlights the need for children and YP to be developmentally involved in decisions across the pathway, for precedence to be given to patients' self-perception and for parents and professionals to be aware of their own values and motivations when facilitating decision making. Clinical implications and suggestions for future research are discussed.

Key words: Cleft, young person, orthognathic surgery, jaw, decision making

Introduction

Treatment pathway

The cleft lip and or palate (CL/P) treatment pathway is long and complex, extending from birth to young adulthood and requiring specialist input from a multidisciplinary team (Alansari et al., 2014; Colbert et al., 2015; NHS England, 2013). Towards the end of the pathway, when facial structures have reached maturation, definitive surgeries may be offered to optimise function and or appearance of the jaw, nose and lip. As unavoidable scarring from primary cleft surgeries can impede typical growth of the jaw and midface (Andersen et al., 2012; Markus and Precious, 1997; Shi and Losee, 2015) patients can present with an underdeveloped jaw and severe malocclusion⁵ (Paradowska-Stolarz and Kawala, 2014; Tang and Lisa, 1992). Depending on severity, this can affect bite and chewing function, satisfaction with facial aesthetics and negatively impact on psychosocial wellbeing (Hunt et al., 2005; Marcusson et al., 2002; Meyer-Marcotty and Stellzig-Eisenhauer, 2009). Orthognathic Surgery (OS) is offered to realign the jaw, create a more typical facial profile and provide a foundation for revision surgery to the nose and lip (Posnick and Tompson, 1995). It is estimated between 25 to 60 per cent of CL/P patients may be eligible for OS (Rachmiel, 2007; Ross, 1987) however it is an elective surgery that patients choose whether to undergo.

As dentofacial maturity is required, OS typically takes place in late adolescence to early adulthood (Robin et al., 2006; Wolford and Stevao, 2002). However, due to the extensive and prerequisite period of preparatory orthodontic work, discussions about OS are initiated earlier around age 16 (Wolford and Stevao,

⁵ Malocclusion can be defined as misalignment or imperfect positioning of the teeth when the jaws are closed

2002). This coincides with the age at which young people (YP) can legally and independently consent to medical treatment, providing they have capacity (Department of Health, 2009; NHS, 2016). It also coincides with the period where YP are developing their identity and sense of self (Erikson, 1963), and as such identification with peers and appearance-related concerns become more salient (Hearst et al., 2010; Wong Riff et al., 2017). Therefore in deciding about OS, YP are required to decide whether a change in facial appearance fits with their developing sense of identity (Aspinall, 2006; Cash and Pruzinsky, 2002). Hence the decision making process and OS itself take place at pivotal point in development; a time where young people are negotiating transition to adulthood, developing independence and autonomy and wishing to have their views heard (Hearst et al., 2010).

Elective surgery and decision making

National healthcare policies in the UK advocate shared decision making to empower patients to feel involved and in control of their treatment, and this includes children and YP (Department of Health, 2010; Coulter and Collins, 2011). Therefore, given OS is elective and takes place when developmentally YP can have a role in decision making, the paucity of research on this topic is surprising. Instead, research has focused upon parents experience of making treatment decisions (Nelson et al., 2012) or children's experiences earlier in the treatment pathway (Hall et al., 2012). Research exploring patients' accounts of undergoing OS or other reconstructive surgeries is limited by gender (Tiemens et al., 2013) and non-specificity to CL/P (Bemmels et al., 2013), however this research does indicate minimising stigma as a key motivation for surgery.

Indeed, when surgery is elective and not medically necessary the factors underlying decisions are inherently complex, with emotional, cultural, social and moral factors playing a key role (Daniel et al., 2005). However current literature does not capture this complexity from the patient perspective in the context of OS. Unfortunately, the studies exploring decision making in OS are not specific to CL/P and do not provide in-depth qualitative accounts (e.g. Rivera et al., 2000; Stirling et al., 2007). Due to the nature of the cleft treatment pathway, it is likely the experiences of YP treated for CL/P will be distinct and thus require investigation. In light of the limitations of their study Hall et al. (2012) advocate for exploration of decision making in the transition between adolescence and adulthood in order to advance understanding.

Objectives

This study aims to qualitatively explore YP's experience of the decision making process for OS in the context of CL/P by answering the following research questions:

1. How did young people with a cleft lip and or palate experience the process of decision making for orthognathic surgery?
2. How can young people with a cleft lip and or palate, and their families be best prepared for, and supported with, making decisions about orthognathic surgery?

Ethical approval

Prior to commencing recruitment, the Health Research Authority granted ethical approval (Appendix 6) and research approvals were received from the two participating clinical centres.

Design

In order to generate rich insights into the experiences of YP, a qualitative methodology was adopted. Qualitative approaches are recommended when working with understudied populations and topics (Creswell, 2007; Flick, 2006; Nelson, 2009) as they are able to respectfully capture the complexity of patients' personal and context-specific experiences and perspectives (Creswell, 2007).

Sampling

Participants were purposively recruited from two specialist, regional cleft centres in England. Inclusion criteria stated participants needed to be 1) a CL/P patient under the care of either cleft centre, 2) a young person aged between 16 and 25 years old, 3) a patient who underwent OS between 6 months and 3 years ago, or a patient who decided not to have OS between 6 months and 3 years ago, 4) English speaking, 5) without an identified Learning Disability (to the extent they would not be able to give informed consent or engage in an interview). The clinical team identified eligible participants and sent postal information about the study.

Interviews

Individual face-to-face interviews were conducted by the principal researcher (MA) between September 2017 to March 2018. Interviews were either held at patient's homes or local cleft centre and lasted between 56 to 115 minutes. Interviews were semi-structured and a topic guide (Appendix 7) was used flexibly to ensure the relevance of data collected. Questions pertained to experiential aspects such as YP's thoughts and feelings when OS was mentioned, the process of deciding, how they felt about making a decision and their readiness as a YP. Participants were also asked about experiences of preparation and support during the process of

deciding. To preserve richness and detail interviews were audio-recorded, transcribed and identifiable details removed with pseudonyms assigned to protect participants' anonymity. All participants provided written, informed consent to participate.

Analysis

YP's accounts were analysed inductively using Thematic Analysis (TA) to yield a rich understanding of their experiences and explore similarities and discrepancies across the data (Boyatzis, 1998; Braun and Clarke, 2006). Analysis was guided by Braun and Clarke's (2006) non-linear, six-phase process for conducting TA. In terms of theoretical positioning, a contextualist, critical realist perspective (Madill et al., 2000; Willig, 1999) was adopted. Reflexivity was employed to consider the impact of researcher subjectivity on the knowledge produced.

Results

Participants

All participants were recruited via the clinical teams as attempts to recruit using the national cleft charity's social media were unsuccessful. Across the two cleft centres a total of 27 patients were identified as eligible to participate and were contacted about the study by their cleft clinical team. Of these, nine did not respond, three declined and three expressed an interest but were unable to participate due to work commitments, geographical location, or because they did not feel ready to talk about their experience.

Interviews were conducted with 12 YP, from across the two cleft centres (7:5). To describe the sample, participants consented to provide the basic

demographic information presented in Table 5. The sample consisted of three females and nine males with a mean age of 21 years 2 months (range 19.5–24.10 years). The ethnic diversity of the sample was limited, with the majority being White British (n=11). Participants were well-educated and were either students (n=6) or employed full-time (n=6). Four participants were born with Bilateral Cleft Lip and Palate (BCLP), four with Unilateral Cleft Lip and Palate (UCLP), one with Unilateral Cleft Lip (UCL) and two with sub-mucous cleft palate. One participant did not know their cleft type. The mean age YP reported deciding about OS was 20 years (range 15-22 years) and participants reported having so far undergone between 1-3 to 10 surgeries. Two of the 12 participants decided not to have OS. The majority (n=10) reported to have met with the cleft Clinical Psychologist at least once, however two participants said they had not.

Table 5. Demographic information for study participants	
Age	Mean = 21 years 2 months Range = 19 years 5 months – 24 years 10 months
Gender	Male = 9 Female = 3
Racial/ethnic background	White British = 11 Asian British = 1
Level of education	A-Level / BTEC = 6 Undergraduate degree = 6
Employment status	Part-time student = 1 Full-time student = 5 Full-time employed = 6
Cleft type	Unilateral cleft lip = 1 Unilateral cleft lip and palate = 4 Bilateral cleft lip = 0 Bilateral cleft lip and palate = 4 Sub-mucous cleft palate = 2 Unknown = 1
Number of cleft surgeries	1-3 surgeries = 5 4-6 = 4 6+ = 3 (range = 7-10)
Age when made decision about Orthognathic Surgery	Mean = 20 years Range = 15-22 years
Decision about orthognathic surgery	Underwent OS = 10 Declined OS = 2
Number of sessions with Clinical Psychologist^a	None = 2 1 session = 5

2-3 session = 2
3-6 sessions = 2
6+ sessions = 1

^aThe national protocol for psychology input to the care of cleft patients who are clinically eligible for orthognathic surgery recommends patients have a pre and post-operative consultation with a Clinical Psychologist as part of the orthognathic surgery process. The availability of psychology is also recommended to support the decision making process or provide intervention as appropriate.

Participants appeared able to speak openly about their experiences, with some being naturally more reflective than others. TA resulted in the development of four key themes: 1) *Awareness of difference*; 2) *Committing to the process*; 3) *Others facilitating decision making*; 4) *Responsibility on my shoulders* (See Table 6). Each theme captures a different aspect of YP's experience of the decision making process and subthemes represent areas of similarity and divergence within themes. Illustrative exemplar quotes are included⁶. A thematic diagram depicts relationships between themes and the multi-layered context of the decision making process (Figure 3).

⁶ Where words from direct quotes have been removed for clarity this is indicated by [...]

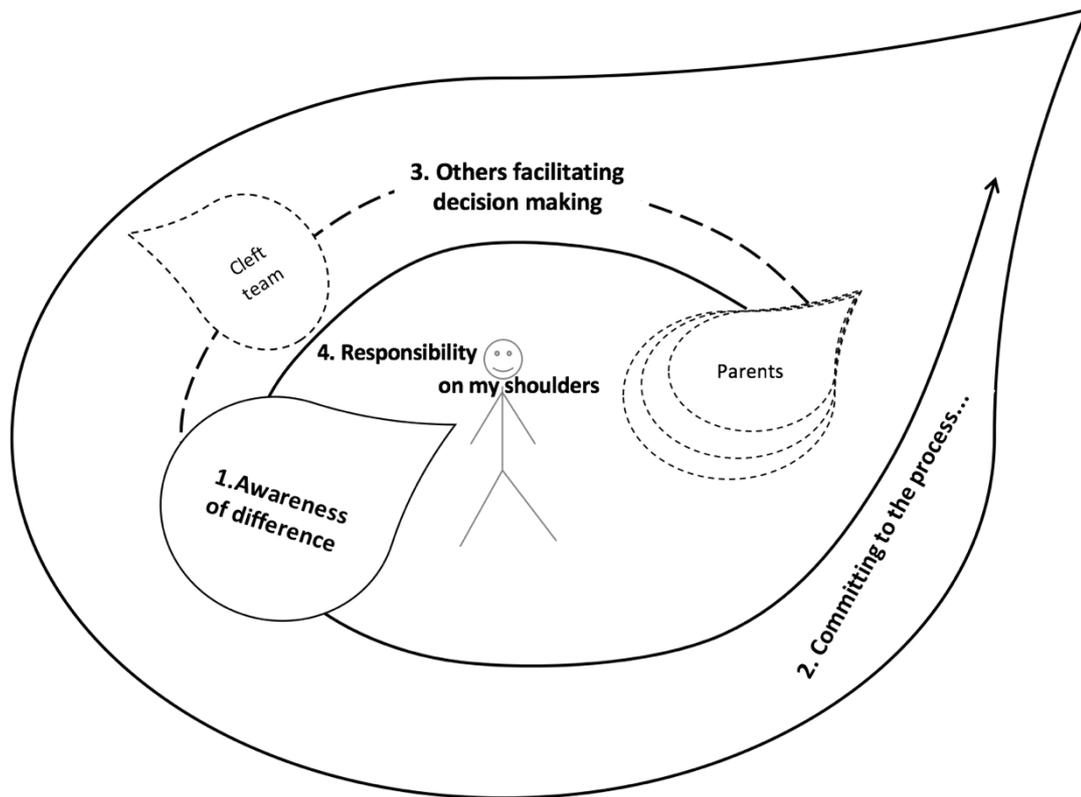


Figure 3. Thematic diagram

Table 6. Themes and subthemes	
Theme	Subthemes
Theme 1. Awareness of difference	<ul style="list-style-type: none"> • Dissatisfaction and desire for change • It's my normal
Theme 2. Committing to the process	<ul style="list-style-type: none"> • It's a long process • Making sacrifices
Theme 3. Others facilitating decision making	<ul style="list-style-type: none"> • Provision of relatable information • Talking it through
Theme 4. Responsibility on my shoulders	<ul style="list-style-type: none"> • Feeling informed, it's up to me • Uncertainty about responsibility • Going along with it

Theme 1: Awareness of difference

YP's narratives contrasted in the degree to which they were aware of, and affected by, the malocclusion before it was discussed or pointed out by their

orthodontist or cleft team. Some YP had an awareness and wanted to change it, whilst others (whether they were aware of it or not) had accepted their jaw as part of themselves. The subthemes '*Dissatisfaction and desire for change*' and '*It's my normal*' aim to capture this distinction.

Dissatisfaction and desire for change. Some YP felt unhappy with their appearance due to their jaw positioning and had themselves already considered further surgery.

I did notice the jaw sort of like sticking out further, the bottom jaw sticking out further, and it was always something that I did notice and it was always something that I wanted to have rearranged, cos I knew that'd benefit me a lot more. (*Tom*)

Others were similarly self-conscious about facial features so the decision about OS was welcomed as it fulfilled a wish to 'improve' appearance through surgery. This underlying dissatisfaction highlights YP's desire to fit in and surgery seemed a route to minimize their sense of difference and improve self-confidence.

I knew I wanted to have it done [...] I thought that I was different from other people because my jaw was different and that I thought that by having the surgery it'd make me feel good about myself (*Amelia*)

I often think if the world was blind, I wouldn't have had half of what I've had done (*Sarah*)

The potential impact of societal pressure to 'look good' is profoundly reflected in both quotes. Evidently these concerns shaped some YP's decision making, especially those concerned about whether their appearance could affect prospects for romantic relationships.

From a self-conscious point of view the thought of it improving looks. I was single at the time, had been single up until that point. I've got a girlfriend now, and I hadn't had a girlfriend previous [...] the looks side of it did make me think, "Well, this would be an improvement, this would help toward that sort of new me" (*Mike*)

It's my normal. Some YP had accepted themselves as they were. Only when the malocclusion was pointed out and OS mentioned did they begin to question this aspect of their appearance.

I kind of got on with the way it was-- and I didn't really think much about getting different types of surgery to improve, it wasn't really an issue for me [...] When you get a choice like that, for example how you look, then you're kind of like actually yeah maybe, maybe I could improve myself, maybe they want me to improve-- to look better (*Patrick*)

It was only really when this, the operation started being discussed by the cleft team that it actually sort of made me aware that, that is how it was, I just hadn't really noticed it on myself and no one had really sort of pointed it out [laughs] (*Mike*)

Terms related to 'correcting' were frequently used in YP's narratives; such language carries the implication of needing to 'put right'. For YP it emphasized being dissimilar to others and introduced doubt about their sense of themselves as 'normal'.

If anything all I remember is the word correcting, that's it. [...] Yeah that's what threw me off cos I thought it was normal [laughs] (*Lara*)

Making a decision about an aspect of oneself that was previously unquestioned was experienced as a dilemma for two participants. One individual had, despite being

bullied, incorporated her jaw as part of her identity so felt understandable uncertainty about undergoing OS.

Cos for a long while even with the bullying I still saw myself as you know me. So then to suddenly want to change that-- do I want to go through that? [...] Cos I thought I was fine. (*Lara*)

Another participant, who in contrast decided not to have OS, articulated that he also experienced it as a difficult decision to make at a time when he had adjusted to and accepted his face as it was.

It was quite difficult to make at that age I guess [...] Just in terms of that I was used to like my face, and how I eat and everything, I was used to it at that age and I didn't necessarily think it was like there was a problem with it [...] personally I just decided-- I was ok how I was, if that makes sense. (*Thierry*)

Theme 2: Committing to the process

Across narratives frequent reference was made to the duration of the decision making process and preparation for surgery and how this commitment affected YP personally and practically, which the subthemes '*It's a long process*' and '*Making sacrifices*' aim to depict.

It's a long process. The process, from the point OS was first mentioned, through deciding and preparatory orthodontics to when YP had surgery, was experienced as lengthy.

I just felt like I was just waiting and waiting for it to happen like-- I think the decision making process was quite a long one. (*Nathan*)

Although some knew about the likelihood of delays due to skeletal growth and readiness of the orthodontics, others had not anticipated a protracted timescale and felt unprepared.

I think the whole making the decision and the whole process of going through the surgery it's quite –it's really long. [...] it would have been nice to know that you know this could take a long time (*Lara*)

Being unprepared meant some YP felt disappointed or frustrated by the timescales involved. However, YP's familiarity with treatment likely enabled them to stay committed to their decision and persist with the process. Indeed, many perceived OS as a treatment milestone they wanted to achieve to feel less burdened by ongoing cleft treatment.

Finishing was a big, big—yeah I think that was really the aim and I wanted to be 18 and have it done, stop having braces, stop being in and out of hospital. Yeah, it was a big finish line of the whole thing. (*William*)

Making sacrifices. In deciding, YP considered the personal impact upon present life priorities, such as sporting interests, work commitments or hopes of going to university. As such, deciding to have OS felt like a commitment and an inconvenience.

I had a bit of a think about it, cos it's like a big thing. It took a lot out of that summer, when I had it done. (*Gareth*)

The only key factors for me that caused me to think about it were being out of action for things like sport, football and things like that. And obviously being out of work. (*Connor*)

Some sacrificed going to University with their cohort, taking a gap year to have surgery. For the two who decided not to have OS their decision appeared partially related to the invasive nature of surgery, recovery period and subsequent impact upon priorities in their lives. For them OS was viewed as an unnecessary inconvenience.

I pretty much wrote it off to be fair. [...] It takes so long to like heal and stuff and I was playing a lot of rugby at the time and I just thought like-- it's just gonna be a pain. Like I just thought it's gonna be a bit of a hindrance, cos it takes so long to repair I won't be able to play sport, won't be able to do like anything physical for ages.
(*Bobby*)

Only because the amount of time of that it like I would have out of sort of normal life and it was, at a time when I think I was like at college and wanting to go to Uni so, I was considering having a gap year just to like leave some time for the treatment, so I thought that that was just quite a big decision to make. (*Thierry*)

YP were concerned about the burden on important others and considered the sacrifices parents would need to make to offer continued support for treatment. For one YP this served to emphasize her sense of feeling different and dependent.

I felt like if I was to go for it, it would be a bit of burden for all the appointments to go to. That sort of thing. So like you're gonna take up part of their time to do stuff for you. Cos I also have a brother and a sister and they've never had to go through that, like they had no appointments. [...] Cos you know all the long driving and the distance and like you know, they also have jobs as well so they need to find the time. (*Lara*)

Theme 3: Others facilitating decision making

This theme illustrates how YP experienced other people (cleft team and parents) involved in the process. Through providing information and discussing options others handed over responsibility and facilitated decision making, albeit to varying degrees. This is captured by the subthemes ‘*Provision of reliable information*’ and ‘*Talking it through*’.

Provision of reliable information. Predictably, information was largely provided by the cleft team, however the extent to which this facilitated decision making varied. YP’s understanding and engagement with the process was affected when discussions were dominated by jargon and technical language.

The only problem that I had in terms of the information given to me-- a lot of it was in sort of professional jargon. [...] they would say it was an ‘oestopathy’ or something like that with lower manular – just loadsa long words where-- you don’t hugely know what they’re talking about. (*Nathan*)

Navigating themselves to a point where they felt adequately informed required YP to have the maturity and confidence to ask for clarification. This was made easier when YP had a personable relationship with a consistent professional as they felt more comfortable asking questions.

At no point did I feel uninformed or misinformed I just felt that I had to ask to feel fully informed, but not because they’re not giving the information but because they’re not giving it in a way that I understood. (*Nathan*)

I knew I could ask [surgeon] whatever I wanted. He’s just a pretty sound guy! [laughs]
[...] I think that is a big thing though, that me and him are like on a level (*Thierry*)

Just as technical information and lack of a relationship with key professionals likely led some YP to feel disempowered, when YP perceived professionals to be preoccupied, impersonal and busy this accentuated the power imbalance. YP felt insignificant and therefore less able to ask personally relevant questions to seek out information that was important to them.

I felt like I couldn't really-- understand and talk to her properly [...] Yeah I spose when they're busy I spose they see so many patients on the-- you're thinking ah she's not bothered but she's probably seen about 10 of me. (*Patrick*)

At the beginning it felt like my worries didn't mean that much to her [...] or whereas if she had taken that time I would have felt more able to speak to her. Kind of even if they're on a time limit making it seem like they're not, sort of builds that trust (*Sarah*)

YP's priority was information about the "relatable effects" of OS; they wanted personalized information about how the surgery was going to affect them, their face and their life.

The clinical stuff-- that doesn't really matter, it just kinda happens, whereas what you really want to know is the less clinical stuff like, [...] your face will look like fuller or whatever rather than saying your jaw would be further forward [...] because a jaw being further forward I've got no real understanding of what that means (*William*)

For some, information provided was framed positively by both professionals and parents, with a focus on discussing the benefits of OS. Some YP found this aided their decision making as it helped them feel reassured they were choosing the 'best' option and perceived they would lose out by not having OS.

Just the word of mouth of the surgeons [...] basically the thing I took away was you'd be missing out by not getting it. All of the positives of it will probably outweigh just

staying and looking the same as you were before. My parents-- they put a little word in saying, "It's not going to do anything but benefit you." (*Gareth*)

Just like the reassurance of everyone around me, sort of like there was never ever anything negative said about the procedure, there was always just positive. (*Tom*)

This focus upon technical aspects and benefits meant a majority of YP who decided to have OS felt underprepared for the aftereffects of surgery and recovery. This topic was one that some YP felt the clinical team "glossed over" or dismissed.

They almost too casually put what like the negative effects straight afterwards would be [...] They did say what would happen they just put it in a way where it didn't sound like it would be a problem. (*William*)

A number of participants mentioned doing their own research to feel better informed and more confident about their decision; this was particularly the case for relatable aspects of surgery and those they felt underprepared for.

Talking it through. Having someone to talk the decision through with was experienced as very helpful by YP, especially when they felt treated as an adult.

They asked me for myself, do I want it done you know they didn't ask my parents that. I feel when you reach a certain age they see you more as an adult. (*Lara*)

When important others, be this parents, the cleft team or the psychologist, encouraged YP to take ownership this facilitated decision making. YP generally felt empowered when they were reminded it was their decision and this contributed to them being able to take responsibility and feel in control.

He [surgeon] was very-- he was supportive as well that it was my decision [...] he was very like this is your choice this is not what your Mum and Dad's telling you to do or your friends it's up to you because you're the one that has to live with it. (*Amelia*)

A few participants valued speaking to the Psychologist as a neutral person who could help them think through the decision and reflect upon how it would affect them personally. Some suggested this aspect was missing from the more "black and white" discussions with the cleft team.

That was the one sort of person or meeting there where it was a bit more-- about me and about how it made me feel and stuff which was nice and, at the time I appreciated that. (*Mike*)

Being given time and space to develop questions, weigh up information and think through the decision was valued by YP as they did not feel rushed into deciding. Often YP used this space to discuss the decision with their parents to help them process information. YP appreciated knowing it was their decision and whatever they decided their parents would support them.

The people that I really lent on were my Mum and Dad, but as I've said they weren't forcing their opinions on me [...] I definitely used my Mum and Dad to like help me understand my own thoughts – I'd kind of say more like as sound boards for how I felt (*Sarah*)

Generally, others respected the YP as the decision-maker and were ready to handover responsibility, although some shared how their parents found this difficult.

Nonetheless, YP were understanding of their parents' inclination to be overprotective given their previously active role in decision making.

I think my Mum probably found it a bit hard just because she'd had such a lot of say for so long [...] it kind of felt like she was trying to get a bit too involved (*Connor*)

My Mum, she very much had an opinion on it, was very much involved in every decision, everything cleft wise before [...] I dunno a part of her probably would want to make the decision for me, is what I'm saying, as any parent would in a caring way. But equally, I think that she was respectful of the fact by that point it was my choice. But she was happy with my choice of saying yeah [laughs] (*Mike*)

For these YP talking it through with their parents was experienced as less helpful and they would have valued more space for their own views and opinions. Having a shared perspective and feeling less isolated in their experience of CL/P was important to several YP and they mentioned wanting to talk it through with similar others. A few sought this out via social media.

There is a girl I actually spoke to [via Instagram] who had the surgery once before me, so I was talking to her and that kind of helped you know, helped me decide that I made the right choice you know—[...] I just found out I wasn't alone you know. I wasn't the only going through this. (*Lara*)

Theme 4: Responsibility on my shoulders

The elective nature of OS was apparent across all YP's accounts. Many referred to the decision as "on my shoulders", however the way they responded to this varied. Some YP felt ready to take responsibility, felt strongly "it's up to me" and wanted to feel informed, whilst others felt uncertain. Some felt tied to the treatment pathway so 'went along' with OS. Whereas others appeared to transition from being uncertain to feeling more comfortable with the decision on their

shoulders. To capture the variation, theme 4 is split into three subthemes *'Feeling informed, it's up to me'*, *'Uncertainty about responsibility'* and *'Going along with it'*.

For the majority, OS signified the first time they felt involved in their cleft treatment or the first time they realised they had a choice. This "big decision" was often their first experience of making a major life decision.

Like this is probably the biggest thing that, well it's the biggest I've had to deal with in my life, for me personally-- I knew that, like I said it was a decision I had to make for myself. (*Amelia*)

So it wasn't really until this operation where I really sort of was involved with it, certainly decision wise cos it was the first one you have - the first one you have that you're involved for the decision (*Mike*)

Feeling informed, it's up to me. Taking ownership was important for many YP as the surgery was ultimately happening to them.

It was my face that was changing and it had to be my decision. (*Amelia*)

Having the responsibility on their shoulders encouraged some YP to take an active role in the process by gathering information and asking questions. YP valued responsibility as it acknowledged their status as adults - able to weigh up information and make a decision about themselves.

The more responsibility I felt that I had, the more interest I took in it. [...] And knowing that you have that responsibility-- is nice (*Nathan*)

I'm an adult and can make these decisions for myself. [...] It felt good knowing that I can make a decision to improve my life or improve my appearance and that I'm 100% involved in that. (*Gareth*)

The decision appeared to sit more comfortably on YP's shoulders when they took steps to feel adequately informed about OS as this enabled them to feel in control of their treatment and ready to decide.

I liked that I was then able to ask for more information, cos I guess that gave me a sense of control that, I was taking control of my treatment and that I knew what was going to happen, that I understood it fully (*Sarah*)

It was common for YP to have understood OS as "a choice of two. I had to get the surgery done or just don't get it done" (*Gareth*). However, in the narratives of those who decided not to have OS this binary distinction was absent. For them, reaching a "well-judged" decision involved exploring and weighing up alternative and additional options for surgery to choose what felt worthwhile.

I was asking about if I left it, then what was there any alternative sort of thing-- are there any other options, or what if we left it what would potentially happen. It probably made it seem a bit better just cos I knew it wasn't a case of like go for the whole jaw surgery-- or nothing (*Thierry*)

Uncertainty about responsibility. For some YP the decision weighed heavily on their shoulders and came as a surprise if they had not anticipated cleft surgery being elective. This emphasises how OS is often the first time YP are expected to be actively involved in their treatment.

I hadn't really expected that I would-- have to make a decision cos I'd had all my decisions made for me, so I think when they-- I kind of got like it presented to me and put on my shoulders I think that's when I was like "oh wow like I gotta make I gotta decide whether I want this" (*Patrick*)

The decision felt like an unwanted burden and perhaps understandably, given their familiarity with decisions being made for them, some YP wanted to defer responsibility.

But it's quite scary when like it becomes your choice, like you want the choice to be made for you [laughs] really. (*Patrick*)

I don't know if I felt qualified enough to make a decision [...] I'd come back thinking like please just tell me - it's better for you if you have or its better for you if you don't have it. (*Nathan*)

Consciously not viewing OS as their decision served to protect one YP from feeling the weight of responsibility on his shoulders and helped to minimise the worry about having OS.

I think it helped as well, not looking at it as a decision cos it's like when you start doubting things and whatnot, then it starts to worry you more. [...] I think if it was put on my shoulders like from the very start sort of like I said, if they kept asking if I wanted it and whatnot then I'd doubt it a lot more. (*Tom*)

In contrast, for others the decision making process became an opportunity to test out their maturity as young adults. Feeling independent in other aspects of life and taking an interest meant some who initially lacked confidence and felt uncertain began to value responsibility and feeling in control.

But then when it comes to the point where you're going to the orthodontist or to the hospital on your own, you think, 'I'm an adult now. I've got—it is my decision to make' [...] yeah there was a responsibility to take an interest and listen. I did feel, I'd say I felt better about it when I started taking more of an interest. (*Nathan*)

Going along with it. Being passive and going along with OS was an alternative coping mechanism. For example, some perceived the discussion about their jaw and OS constituted a recommendation for surgery, even if the optional nature was outlined.

I don't want to say he recommended it, but it was that thing-- like, "You're not really going to lose anything by going through it." That was probably the motivating factor, and then just didn't really think twice about it, just go for it [laughs]. (*Gareth*)

Trusting the cleft team were mentioning OS for a reason was common, and a route to deciding for several YP who placed their trust in professionals to act in their best interests.

Obviously they knew what they were doing and they were highly qualified so I knew they knew what they were talking about and I knew they had my best interests at heart so (*Connor*)

No I sort of took what they said as gospel really. I put my whole trust in them. [laughs]
I always just thought there's a reason that the medical professionals are recommending it so I should do it (*Nathan*)

Some YP admitted to not feeling ready to make a decision and described 'going along with it' by agreeing at various stages of the process leading up to OS.

I kind of feel like I was never really ready to make a choice. You kind of, it kind of just happened I guess, like I said earlier it wasn't like a time where I was like 'yes' it was kinda like had the meeting then it flowed into brace-work with still the option open and then [...] you know when you kind of just do something and it's happening you're just kinda doing it. (*Patrick*)

You say kind of yes to a little thing and then yes to another thing, so until you look back at it, none of them were that big. (*William*)

Two other individuals did not consider OS a genuine option, instead it constituted a routine, and therefore unquestioned, part of cleft treatment and one reportedly normalised by the cleft team.

I guess I've not really thought about the idea that people don't have this, I've always thought yep cleft lip –you're gonna have jaw surgery [...] I think at the time, it felt very sort of routine and that was the path [...] just it's part of procedure so, this is what other people have done (*Sarah*)

It was always in my head it was just gonna to happen. But I always looked at it as not a decision, like it was just going to happen with the cleft and whatnot. Yeah, just sort of like, what most people with cleft lip and palate go through. (*Tom*)

Discussion

Summary

This study offers a first insight into the experiences of YP making a decision about undergoing OS, in the context of the cleft treatment pathway. The findings go some way to reveal the complexity of the decision making process; a process within which YP are required to balance their wish to end the burden of treatment, with their own sense of personal and social identity and current life priorities as well as the expectations and opinions of authority figures. For YP at a transitional point in their development, deciding about OS represented a major decision and key milestone in terms of making independent decisions and becoming an adult.

Key findings

Thematic analysis of YP's accounts revealed differences in how they

responded to the decision ‘on my shoulders’. Individual differences in the extent to which patients wish or feel able to be involved in decision making are also recognised in the literature (Coulter, 1999; Hall et al., 2012). Nonetheless all but one participant felt strongly about being involved and making their own decisions, which is perhaps attributable to this being a young and educated sample (a social group known to want to take an active role; Coulter and Collins, 2011). Aside from the ethical imperative of patient involvement (Coulter and Collins, 2011; Department of Health, 2010), the need to involve YP in decisions about them is confirmed by evidence indicating youth with craniofacial conditions appraised their surgical outcomes positively, and reported higher levels of satisfaction when they were involved in deciding (Kapp-Simon et al., 2015).

In our study, YP’s involvement and ability to make an independent and informed decision appeared affected by the extent to which others relinquished responsibility and put aside their own opinions to facilitate YP’s decision making. This is similar to another craniofacial study where parental influence impacted upon decision making (Bemmels et al., 2013). As the motivations of both professionals and parents affect the way information is presented and perceived, it is suggested others involved in the process are aware of their own values (Aspinall, 2010). This will enable others to better support YP by facilitating balanced discussions about all treatment options, without imposing their own views.

An unexpected finding was that only those who decided not to have OS appeared informed about alternative options. Ensuring awareness of all available options (including the option of no further treatment) is essential to facilitating informed decision making (Coulter and Collins, 2011), especially in the context of

the cleft pathway where YP's relationship to medical authority and the expectation of ongoing treatment may impede decision making. This was evidenced to some extent by the theme '*Committing to the process*' where YP made personal and social sacrifices to undergo OS as the milestone to end the burden of treatment. This resonates with another study which highlighted how patients undergoing OS were bound to the lengthy process (Cadogan and Bennun, 2011).

Although YP talked about 'informed' decision making what YP understood by this or the extent to which they engaged in it was not formally explored. Some weighed up information, which is considered a key aspect of informed decision making (Bekker et al., 1999). However, in real-world situations truly informed decisions (made without the use of a decision aid) are considered unlikely because to cope with cognitive load we engage in heuristic processing to attend to and simplify certain aspects of information (Chaiken, 1980). Use of heuristics was apparent for YP in this study in the way they prioritised attending to information with a positive bias, simplified the decision to a binary choice or were influenced by who made the 'recommendation' (Marewski and Gigerenzer, 2012).

Evidently being informed was only part of the picture as contextual factors played a key role in YP experiences. To make shared decisions it is evidenced that patients need not just knowledge but power – that is an awareness they can influence the process (Joseph-Williams et al., 2014). 'Going along' with OS either due to it being perceived as a recommendation or routine part of treatment could imply a perceived lack of power. Alternatively, some consider that deferring responsibility is a decision in itself (Mårtenson and Fägerskiöld, 2008) and one that communicates a trust in others to act in their best interests, which resonates with paternalistic models

of healthcare (Coulter, 1999). Developmentally involving children and YP in decisions (Alderson and Montgomery, 1996) across the cleft treatment pathway would give patients confidence that their opinion matters, and help them feel prepared to take on increasing amounts of responsibility, until they feel ready to be the primary decision-maker.

A barrier to engagement and feeling informed was the provision of technical rather than personalised information, and these views were echoed by participants in another cleft study (Alansari et al., 2014). Undoubtedly, the provision of age-appropriate and accessible information is a necessity to permit children and YP the chance to be competent decision makers (Mårtensson and Fägerskiöld, 2008). Our study also illustrates that professionals can best support YP to engage in decision making by working in partnership to build personable relationships and offering information that matters to YP, thereby aligning with the ethos of shared decision making (Coulter and Collins, 2011).

Prioritising the patient perspective seems imperative as individuals affected by CL/P often perceive themselves differently to those around them (Bemmels et al., 2013; Brattström et al., 2005; Meyer-Marcotty and Stellzig-Eisenhauer, 2009; Semb et al., 2005; Sinko et al., 2005). A presumed positive self-perception was apparent for some YP in this study where the mention of malocclusion and OS came as a surprise and jarred their sense of self. A study exploring adolescent girls experiences also found patients only began to doubt their jaw profile after it was pointed out (Tiemens et al., 2013). Comparably, other research notes, as this study does, a distinction between those actively wanting surgery and those who consider it due to perceived external pressure (Bemmels et al., 2013; Kapp-Simon et al., 2015).

Perhaps in our understandable motivation to achieve the ‘best’ clinical outcome we may be inadvertently reinforcing society’s stereotype of facial attractiveness (Cunningham, 1999). This raises an ethical question as to whether in offering OS we are attempting to ‘alter their normal’, by encouraging individuals to question their identity at an already turbulent period of development.

With this in mind this study raises the question as to when is the best time for YP to make a decision about appearance altering surgeries? The majority, but not all, YP felt ready to make a decision, with support. One might be inclined to agree with this based on the beginning of the decision making process for OS typically coinciding with the development of higher cognitive abilities (e.g. hypothetico-deductive reasoning, meta-cognitive skills and moral reasoning). However, depending on life experience, YP might lack the capacity to fully grasp the long-term consequences of their decision until around age 21 (Partridge, 2014; Partridge, 2010) and compared to adults YP are thought to have greater difficulty placing their decision in a ‘temporal context’ (Piker, 2011). Moreover research suggests that even without appearance-altering surgery self-acceptance increases over time, especially after age 18 (Zuckerman and Abraham, 2008), likely because peer influences and social pressures wane, values develop and priorities change. This finding was replicated in narratives of adults treated for CL/P who reported that with age, many had become more accepting of residual CL/P features (Stock et al., 2016). However, it is acknowledged that these adults are from a different generation who were not privy to the centralised model of specialist care or surgical advancements, so whether YP in today’s society will also experience an increase in self-acceptance over time is uncertain.

Critical evaluation

Many agree that to deliver effective and sensitive healthcare we need to first understand the patient perspective (Aspinall, 2010; Donabedian, 1996; Nelson, 2009). A strength of this research was a commitment to exploring YP's experiences, a group often excluded from craniofacial research (Hunt et al., 2005; Mouradian et al., 2006). This study was also the first to explore decision making about elective CL/P surgeries (Hall et al., 2012; Hunt et al., 2005). Furthermore, the qualitative methodology allowed for in-depth insights, and the additional perspective offered by the two individuals who decided not to have OS is valuable in highlighting possible differences in priorities, adjustment and self-perception.

More participants were male however this reflects more males meeting inclusion criteria. Recruitment via clinical teams meant the sample was limited to those still receiving care, and there was a sense that participants were keen not to be critical. Given that all participants appeared pleased with surgical outcomes there may also have been a bias when reflecting on their experiences, and the study is limited to understanding the experiences of those who participated (who may have experienced the process differently). Nevertheless, findings still offer much needed insight into the unexplored perspectives of YP deciding about OS.

Clinical implications

The unexpected finding that some YP were oblivious to or unperturbed by their jaw suggests a need for greater sensitivity in how OS is discussed, especially given the power dynamics and nature of the cleft pathway. Prioritising patients' self-perception by assessing self-reported psychosocial adjustment and satisfaction with appearance before discussions are initiated would help to inform this process. This

may also identify the need for psychological intervention to resolve underlying difficulties⁷.

Further research

To gain a complete understanding of the decision making process from all perspectives future research should aim to capture parents' and professionals' experiences. Given both counterparts previously held more responsibility and are key to facilitating decision making it seems pertinent to explore their perspective. As some participants spoke about gaining confidence or accepting themselves post-surgery it would be worthwhile investigating whether self-concept increases after OS in CL/P. Exploring possible differences in self-perception between those who had OS and those who did not would also be beneficial.

Conclusions

By exploring the experiences of YP this study reveals some of the contextual, social and personal complexities involved in deciding whether to undergo an appearance-altering surgery during a key period of development and transition. It highlights the need for children and young people to be developmentally involved in decisions across the cleft pathway and for precedence to be given to patients' self-perception.

⁷ Further clinical and practice implications are detailed in the extended discussion, Chapter 5.

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Chapter Four: Extended methodology

This chapter provides an extended outline of the methodology employed for the empirical study to supplement information already detailed in the paper. Further explanation is provided about the design, selection of participants, method of data collection and ethical considerations. An overview of thematic analysis is provided including consideration of the researcher's theoretical positioning and subjectivity, in addition to a reflection on the quality and rigour of the present study.

Rationale for qualitative design

Considering the study's aim to explore and understand the experience of decision making from the patient perspective a qualitative methodology was chosen. Unlike quantitative methods, qualitative approaches can capture the complexity inherent within people's subjective experiences and they enable us to enhance our understanding of people's social and psychological worlds (Braun & Clarke, 2013; Elliott, Fischer, & Rennie, 1999; Polkinghorne, 2005). Remarkably, compared to other fields of health research there are relatively few qualitative studies within the field of craniofacial research (Nelson, 2009). Moreover, a key criticism of craniofacial research is the lack of studies exploring the patient perspective (Stock, Feragen, Moss, & Rumsey, 2018) meaning the subjective experiences of affected individuals are often excluded (Hunt et al., 2005; Mouradian et al., 2006). Use of qualitative methods has therefore been encouraged in this field to develop understanding about patient values and experiences, to ensure services support patients in the ways that matter most (Nelson, 2009). In addition, recruitment of large samples is not always possible meaning application of qualitative methods are well-suited to this specialist field (Stock et al., 2018).

Selection of participants

As is typical in qualitative research, purposive sampling was employed to recruit YP with specific experiences relevant to the study objectives (Patton, 1990). In order to capture the spectrum of experiences, efforts were made to recruit patients who had both made the decision to undergo OS and patients who decided not to. It seemed important to recruit those who had decided to not undergo OS as their perspective is not present in the literature and other research has been unsuccessful in recruiting them (Nelson, Caress, Glenny, & Kirk, 2012).

Due to the idiosyncratic nature of much qualitative research there are no clear rules about what constitutes an adequate sample, instead this depends on the aims of the research and topic under study (Morse, 2000; Patton, 2015). To provide a rich enough dataset to undertake meaningful Thematic Analysis (TA), and allow for potential variation in the richness of data (Braun & Clarke, 2013; Morse, 2000), the present study chose a sample size of 12. This decision was guided by research which explored how many interviews are needed in qualitative research and concluded that data saturation⁸ tends to occur within 12 interviews but suggested pattern repetition was also evident in just six interviews (Guest, Bunce & Johnson, 2006). This was similar in the present study where saturation, and recurring patterns of meaning were apparent after 10-11 interviews. As the intention of qualitative research is to develop understanding, and to link findings to theoretical knowledge, rather than be generalizable or representative of a population, a large sample is less important.

⁸ Saturation is a concept borrowed from Grounded Theory and is a term used to describe when enough data has been collected based on the observation that additional data does not generate new meanings (Morse, 1995).

In line with the recently updated Data Protection Act and General Data Protection Regulation (GDPR; Information Commissioner's Office, 2018) the cleft teams at each hospital were responsible for identifying eligible participants from the patient databases. Clinical teams sent an 'initial contact pack' providing information about the study to all eligible patients. This included the invitation letter (Appendix 8) recruitment advert (Appendix 9) and participant information sheet (Appendix 10). Patients interested in learning more about the study were invited to express their interest by completing and returning the consent to contact details reply slip or contacting the researcher directly. To facilitate recruitment, the clinical team made telephone contact with patients who had not responded to establish their interest in the study. This strategy was discussed with the clinical and supervisory teams and the cleft patient representative and was deemed a helpful way of facilitating recruitment, particularly as patients are typically very engaged with their cleft team. To optimise recruitment, the study was also advertised via the national cleft charity's (Cleft Lip and Palate Association; CLAPA) regional Facebook pages (linked to each of the clinical centres involved in the study) and CLAPA Twitter account. Both the clinical team and patient representative believed it was important to use these online forums to highlight the research to YP.

Those who expressed an interest in participating were provided with further details about the study and their questions answered. For those who volunteered to participate a time, date and location for the interview was arranged. On the day of the interview participants were reminded of the research rationale, purpose of the interview, and their informed consent was obtained (Appendix 11). A letter was sent to the participant's GP to notify them that their patient had taken part in the research. Participants were given a £10 voucher as a small token of appreciation for their time

and participation. Contact details of the researcher were provided in the event of questions or concerns.

Data collection

Interviews were chosen as the most appropriate method of data collection in order to generate rich narratives of YP's experiences (Braun & Clarke, 2013; Green & Thorogood, 2014). Use of a topic guide (Appendix 7), developed in collaboration with the research and clinical teams and CL/P patient representatives, ensured the relevance of data collected by asking participants broadly similar questions about their experiences. However the guide was used flexibly, with each interview being tailored according to participant's developing narrative (Braun & Clarke, 2013). As recommended, questions began broadly and became more specific as the interview progressed and rapport was established (Braun & Clarke, 2013; Kvale & Brinkmann, 2009). The open-ended nature of questions and use of probing and follow-up questions enabled participants to speak about experiences salient to the decision making process.

All interviews were face-to-face as despite advances in technology this approach still seems to be viewed as the optimal way to collect interview data (Novick, 2008). It was also the preferred method for both YP and the researcher although the option to conduct telephone interviews was available.

Patient and public involvement

Involvement of patient representatives in research is recommended to ensure quality research that has relevance for the population being studied (Brett et al., 2012). Accordingly, a panel of patient representatives at one of the hospitals was consulted about the study and invited to provide feedback on the research design,

including the recruitment strategy, and to comment on research materials such as the participant information sheet, demographic information form (Appendix 12), consent form and interview topic guide. Feedback received from the panel review was positive, commenting that they found the research materials “user-friendly, detailed and sympathetic”. Suggestions for minor changes included grammatical changes and altering the phrasing of the term “orthognathic” to “corrective jaw surgery (orthognathic surgery)”. It was felt altering this phrasing on the study documents would ensure participants were not daunted by medical terminology. In addition, the panel also recommended further clarity in the participant information sheet regarding the potential impact of the study, however they agreed it was a “well-considered and important” study.

A regional CLAPA coordinator supported the study and enlisted the expertise of a CL/P patient representative who advised on the recruitment strategy and reviewed research materials. The patient representative also took part in a pilot interview to inform the interview process by enabling the researcher to consider how questions were received and answered to ensure the richness of data collected.

Ethical approval and considerations

The research proposal was subjected to internal review at the University of East Anglia and approved as a suitable research project. Formal ethical approval was granted by the London Brent Research Ethics Committee (Appendix 6). Approval was also granted by the Research and Development departments in the participating clinical centres.

Although no significant ethical, legal or management issues were raised in the design or conduct of this study, there were a number of relevant ethical

considerations to ensure the research conformed to the Code of Human Research Ethics (The British Psychological Society, 2014) and the recently updated Code of Ethics and Conduct (The British Psychological Society, 2018).

Eligibility criteria. Only YP within the defined age range of 16-25 years (inclusive) were eligible to participate. This age range was chosen because decisions about OS are most likely to occur during this period. There is also limited research exploring the direct decision making experiences of YP within this age period, this is despite 16 being the age when individuals can legally and independently consent to, or refuse, medical treatment.

Only YP who underwent, or decided not to have, OS between 6 months to 3 years ago were eligible to participate. These criteria were set based on discussions with the surgical team to ensure those who had the surgery had a sufficient recovery period (e.g. at least 6 months) before being contacted.

Unfortunately, those unable to speak fluent English or who had an identified learning disability were excluded from participating. This could be considered an ethical issue as people should be given equitable access to take part in research and it means the views of these groups were not adequately represented. However, as there were no funds to pay for interpretation (owing to the fact this research was undertaken as part of a doctoral course) this is noted as a limitation of the study. The decision to exclude those with an identified learning disability was reached after considering that their involvement in the process of decision-making might be quite different from those without; in addition the study required participants to reflect on their experiences. It is also important to note that qualitative research does not aim to

be generalizable, though future studies may wish to focus upon inclusion of these individuals.

Informed consent. The clinical team obtained patients' initial consent for the researcher to contact them about the study. Patients were provided with the participant information sheet prior to the interview to ensure they had at least 24 hours to read the information and develop questions. To ensure readability and provision of adequate information the participant information sheet was drafted, and feedback sought from patient representatives. One participant provided spontaneous feedback that she knew what to expect from interview as the participant information sheet was clear.

Informed consent to participate was obtained on the day of interview by ensuring questions were answered and that participants understood the purpose and nature of the study. The researcher monitored participant's ongoing consent throughout the interview, and participants were aware that during data collection and for one week after the date of the interview they could modify their consent or request for withdrawal of part or all of the data provided.

Coercion. Issues of coercion were minimised as the clinical team made initial contact with eligible participants by sending the initial contact pack and by conducting follow-up telephone calls. Interested participants gave their consent for the researcher to contact them about the study. All participants were made aware that by providing consent to contact they were under no obligation to participate in the research (this is evidenced by three YP deciding not to participate due to other commitments). Participation was therefore voluntary and participants were aware

they could withdraw from the study, without providing a reason, and that this would have no effect on the care received from their cleft team.

Confidentiality. Interviews were confidential. Personally identifiable information was removed from transcripts and a pseudonym assigned to protect participants identity. Personal information and data were handled sensitively in accordance with the principles of data protection and the GDPR (Information Commissioner's Office, 2018).

Disclosure. Participants were made aware prior to commencement of the interview (both in the participant information sheet and when providing informed consent) that any disclosure (such as risk to self or others) would result in a confidentiality breach to an identified clinician in order to ensure the safety of individuals concerned, however any breach would be discussed with the participant.

Deception. No deception was involved as participants were given full details about what the study involved and how the information they provided would be used.

Distress. There was a small possibility that participants could have become distressed when sharing their experiences during the interview, given the personal nature of the topic. This was mitigated to some extent by the interviewer having professional training and experience in managing distress. All participants received details of support agencies at the end of the interview, and had it been necessary participants would have been advised to seek further support from the cleft team, GP or alternative agencies.

Risks / burdens / benefits. The risks and burdens posed to participants were minimal and it was felt they may even benefit from the opportunity to reflect on their experiences. Participants took part in a face-to-face interview and did not receive any form of intervention. To minimise travel and financial burden, interviews were held at a convenient location for the YP (at their home or local cleft centre). Participants were given a £10 voucher as a goodwill gesture for participating. To ensure researcher safety and minimise risks when conducting interviews, the lone working policy was adhered to and the ‘safety buddy’ procedure adopted.

Thematic Analysis

Thematic Analyses (TA) was chosen as the method of analysis as it focuses upon locating recurring patterns of meaning across a dataset (Boyatzis, 1998; Braun & Clarke, 2006). TA also places emphasis on situating experiences in a social-cultural context which seemed relevant in view of decision making about appearance altering surgery being the topic under study. A decision was made to conduct TA inductively using a data-driven approach to derive themes due to limited understanding of YP’s experiences of making decisions about elective cleft surgery. Given the exploratory nature of the research, the analysis also aimed to provide a rich but broad description of the entire data set (Braun & Clarke, 2006). It was assumed there would be some overlap in YP’s experiences therefore TA was considered the most suitable method to meet the aims of the present study, when compared to other qualitative methods that either focus upon individuals (Interpretative Phenomenological Analysis; (Smith, 1996) or aim to generate a theory (Grounded Theory; Glaser & Strauss, 1999).

Analyses was guided by the recursive, six-phase process for TA (Braun & Clarke, 2006). As a first step in the analysis, notes were recorded after each interview with the researcher's initial reflections and ideas (Maharaj, 2016). Phase one involved transcription and familiarization with the data. Interviews were transcribed orthographically, re-read and checked for accuracy. To immerse oneself in the data transcripts were read actively a number of times and initial ideas noted. This was followed by systematic coding at the semantic level (phase two) then sorting and collating of coded data extracts with related concepts to identify patterns/themes (phase three; see Appendices 13 & 14 for example of a coded transcript and example coding and theme development respectively). Suitability of candidate themes were checked by referring back to collated data extracts. Themes were discussed and refined as needed (phase four) to ensure the salience and meaningfulness of data captured within them (internal homogeneity) and to ensure adequate distinction between themes (external heterogeneity; Patton, 1990). A thematic diagram (Figure 3, Chapter 3) was also created to evidence the construction of and relationship between themes. Themes were created based on the contribution they made to understanding YP's experience of decision making rather than how often they occurred in participants' accounts. Themes were further defined and named (phase five); where possible themes were named using participants language. In the final analysis (phase six) the most vivid quotes were selected for inclusion to illustrate key themes and to demonstrate how data were analysed and themes derived (Braun & Clarke, 2006; Sandelowski, 1994).

Theoretical framework and positioning

Being the most flexible of the qualitative approaches, TA is not attached to a specific theoretical framework (Braun & Clarke, 2006). As theoretical positioning

affects all aspects of research from conception to final conclusions transparency is important. In terms of an epistemological framework, concerning the nature of knowledge, the researcher aligned with a contextualist perspective (Madill et al., 2000). This assumes that knowledge is situated in and affected by context so there are multiple perspectives rather than a single, universal truth. In terms of ontology, concerning the nature of reality, a critical realist stance was adopted (Willig, 1999). This acknowledges that reality can only be partially accessed as it exists through people's perspectives, and as such only the experiences reported by participants could be analysed. The critical realist stance also considers how the societal and social context can influence the sense we make of experiences.

Researcher subjectivity and reflexivity

In qualitative research the subjective influence of the researcher contributes to the design, methodology and research outcomes. Considering the researcher's influence on the knowledge produced (reflexivity) enables inevitable researcher bias and subjectivity to be utilised as a meaningful part of the research (Koch, 2006). To facilitate reflexivity, and consider my influence on the research process, I maintained a reflexive research journal. For example, following each interview field notes were made to record my initial analytic thoughts about the data. During analysis I allowed time to consider how I was reading the data in relation to my own experiences, values and beliefs, and being aware of this meant I was to some extent able to bracket these reflections (Elliott et al., 1999). However it is acknowledged that the data itself were produced via the interaction between the participant and myself as the primary researcher (Polkinghorne, 2005).

Part of reflexivity involved considering how my own personal and professional values, beliefs and experiences likely impacted upon the knowledge produced from this study and these are outlined below.

Personal reflexivity.

Choice of research topic. My choice to undertake this research was influenced by a number of factors. Firstly, I was intrigued by CL/P as a condition and the nature of the developmental treatment pathway extending from birth to adulthood. Being a psychologist meant I was curious about the process involved in deciding whether to have surgery to alter your appearance. I wondered whether YP genuinely feel like they have a choice about elective surgery, within the context of the medical, family and societal setting where they may feel disempowered. Ethically, I was struck by the idea of so called ‘normalising surgeries’ which aim to minimise difference.

Personal experiences and background. Reflecting upon my own cultural background and upbringing, I am a 28-year old White British female. Growing up I was encouraged to make autonomous decisions, though with my parent’s guidance and support. My age, cultural background and experiences of being parented and developing independence likely affected the way I interpreted and understood the data.

Surgical and orthodontic experience. In my early childhood I had non-elective surgery on my chin which left a scar and clinicians advised my parents that, depending on how it healed, they may wish to consider plastic surgery at a later stage. This was not something we ever pursued as my scar healed and just became part of who I am. Reflecting on this at the outset of this research led me to consider

how a decision about plastic surgery might have been reached and the possible extent of my involvement in it.

In terms of orthodontics I elected to have braces fitted in adolescence but I remember being swayed by the views of my parents and the orthodontist. To the contrary my sibling chose not to have braces. Despite these experiences I do not have a CL/P, nor do I know personally of anyone affected by CL/P. Therefore, aside from reading literature and engaging with the cleft team and patient representatives, I had limited awareness at the outset of this research of what being affected by CL/P is like. To enrich my understanding of participants' worlds I observed some cleft clinics and engaged in discussions with the cleft team in the hope of developing more nuanced insights into YP's realities.

Influence of being a Clinical Psychologist in training. It is likely that my training as a Clinical Psychologist influenced the research process in terms of the study design, data collection and analysis. Such professional training and experience could be considered to have had an enhancing or equally a biasing influence on the research process and outcomes. For example, my training in understanding human cognitive and emotional processes and behaviour, in addition to experience of measuring and evaluating PROs could have enriched this thesis due to having nuanced insights. Whilst it is acknowledged that my training and experience could have made me oblivious to certain aspects within the data it is hoped the inductive, data-driven approach to analysis, reflexivity and regular research supervision would to some extent have helped to mitigate such bias.

Furthermore, attendance at the University qualitative research forum helped me to consider the importance of switching into the researcher role when conducting

interviews. This meant I became more aware of gathering information to answer my research questions and utilised clarifying questions to ensure I had understood the participants perspective. It is possible that my clinical experience of working therapeutically with clients and adopting an empathic and non-judgmental stance could have enhanced data collection by enabling participants to feel at ease and able to talk about their experiences. Additionally, my training meant I could make use of conversational pauses in the interview which allowed time for the participant to naturally expand on their narrative, often without additional prompting.

Conducting this research has impacted upon my clinical practice as I feel I have developed better skills in being able to actively listen to client's narratives and feel more confident in tolerating moments of silence in conversation to illicit additional information.

Functional reflexivity.

Interest in health and paediatric psychology. I have a longstanding interest in health and paediatric psychology, stemming from previous roles in physical healthcare settings focusing upon patients' experience of care. This led me to develop beliefs about the importance of empowering patients to be active participants in their healthcare, in order to optimise both their psychological and physical wellbeing. I am also interested in the differences in the way paediatric and adult care is delivered and how patients can be best supported to transition between services. As cleft teams tend to view themselves as lifespan services the issue of transition and increased patient responsibility seems pertinent.

Interest in qualitative research. In terms of my influence on the methodology I tend to align more with qualitative research as it is able to capture complexity, provide privileged insights into participants' worlds and prioritises the

voices of affected individuals through the interpretation of their narratives and experiences.

Quality and rigour

Various guidelines exist by which to measure quality in qualitative studies (e.g. Elliott et al., 1999; Yardley, 2000) These were largely developed to permit fairer reviews based on criteria relevant to qualitative methods rather than borrowing standards from quantitative methods, which tend to adopt a positivist perspective at odds to much qualitative research. Some of the key markers of good qualitative research the present study aimed to meet were paying attention to matters of subjectivity (Elliott et al., 1999), sensitivity to context (Koch, 1994, 2006; Yardley, 2000, 2017) and illustrating with examples of quotes (Elliott et al., 1999). In terms of credibility checks, regular research supervision enabled discussion of the research process including the development of codes and themes to ensure an inductive approach. Attendance at the University qualitative research forum also permitted discussion of qualitative methodologies to improve conduct of the research.

Braun and Clarke (2006) put forward a 15-point checklist for ‘good thematic analysis’. This was borne in mind during data collection and analysis to try to ensure a rigorous approach. For example, data were carefully transcribed and checked against recordings for accuracy, the extracts used aim to clearly depict the analytic ideas and the active role of the researcher is evidenced, such that themes ‘emerged’ not from the data but from the researcher’s interpretation of and interaction with the data (Ely, Vinz, Downing, & Anzul, 1997).

Chapter Five: Extended results and discussion

Extended results

Due to word limitations of the chosen journal, the results section of the empirical study details a broad overview of the central findings. Therefore, this section aims to present supplementary findings and considerations not captured within the thematic analysis. To further substantiate the themes developed additional participant quotes are presented in Appendix 15, as only the most vivid quotes and participant extracts were included in the empirical paper (Braun & Clarke, 2006; Sandelowski, 1994).

Influence of gender and culture

Apparent across YP's narratives was the potential influence of gender, peers and cultural background on YP's experience of the decision making process. In particular it was noted that males spoke less than did the three females about the influence of stigmatising experiences on their decision, instead, their accounts imply they experienced peers as accepting or oblivious to cleft-related features. This finding is akin to other research where males with facial difference report less stigma than females (Strauss et al., 2007). Whether there is a genuine difference or whether males are less likely to acknowledge and discuss stigma experiences is unclear. Similarly, although no conclusions can be drawn, it was observed that the two YP who decided not to have OS were both male.

An individual's cultural background, in terms of their beliefs about interacting with authority figures, also appeared to affect how YP experienced the decision making process. For example, those who believed 'doctor knows best' or 'I should do what my parents say' had narratives which indicated that they found it

harder to ask questions and share their own opinions, unless it was explicitly encouraged by authority figures.

Participant feedback on the interview process

Several participants commented upon the value they derived from sharing their experiences. For some, it was a first opportunity to reflect upon and consider their experience from a different perspective and they were glad to have participated.

I've spoken bits about it at length but not that bit so it was weird thinking about stuff I've never considered as much. But quite nice to get it and let some of it out there. No it was good. I'm pleased I did it. But no I quite enjoyed it, nice to talk about it.
(William)

For one YP taking part in the interview helped her to feel less alone in her experience of being treated for cleft and making decisions, as she recognised that other people had also decided. Through being encouraged to talk about her experiences she was able to recognise the importance of disclosing her values and opinions.

If anything this interview is probably the most I talked about-- from my side, cos it's the sort of thing I normally hide. It's been nice knowing you know, it's not something to be closed about, it's an open thing. Other people are making the same decision you know. It's not something that's like on your own shoulders [...] You're not the only one. *(Lara)*

Inviting YP to participate in this research communicated the message that we are interested in their experiences and value their voice. For those who felt unable to participate and share their experiences this may have in itself been helpful to realise there are other YP going through a similar process. This is important because YP's

narratives revealed a sense of feeling alone in their experience of cleft treatment and in making a decision. A large proportion of YP explicitly commented on the potential value of having peer support to gain a shared perspective on the decision making and surgery processes. Others, through searching for peer perspectives online, implicitly suggest this would be a helpful addition to the process. Primarily YP wanted to speak to similar others to feel more prepared for surgery, recovery and coping with the after-effects.

I feel like talking to someone who'd actually had it done before, would have just benefit my, or benefit what I think was going to happen during the operation, and just help me to have a clearer picture of what I'm gonna be like afterwards. What's going to happen during the operation, the recovery time, how it's going to affect me, stuff like that. (*Gareth*)

However, peer support and interaction could be multi-purpose, for example, by reducing the sense of isolation felt by some being treated for CL/P, by presenting an opportunity for YP to ask the "silly questions" they may not feel able to ask the clinical team, and to potentially hear from YP who decided not to have OS.

Researchers in the field advocate peer support (Stock et al., 2015) likely because it is considered beneficial to both those providing and receiving support. A number of YP in the present study expressed an interest in supporting others by sharing their experiences.

Recruitment challenges

In recognising that the views of affected individuals are often excluded from craniofacial research, Nelson (2009) advised researchers to engage YP in qualitative research to develop our understanding about issues directly affecting them. Although

this study fulfils this recommendation, a number of challenges were encountered in recruiting YP. The eligibility criteria and recruiting from a relatively small medical population such as CL/P meant the pool of eligible patients was quite small, even across two clinical centres.

To aid recruitment, posters were displayed in the two participating cleft centre waiting areas though no one expressed an interest from these. Although both the patient representative and cleft team felt recruiting via CLAPA social media was important to allow an opportunity for individuals to self-select this was unsuccessful too, with no one expressing an interest via CLAPA. This could be because in order to see the study advert YP needed to first have a connection with CLAPA and then needed to be motivated to express their interest directly. In a study exploring adults narratives (Stock et al., 2016) the authors successfully recruited half of their 52 participants via CLAPA. This could suggest that more direct approaches are needed to recruit YP.

The 12 who participated were all recruited by the clinical teams. Of these, three returned the consent to contact form without additional follow-up, which suggests a motivation to participate and share their experiences. However, follow-up was required to achieve the overall 44 per cent participation rate. Clinical teams telephoned all those who had not responded to ascertain consent to contact. Several people requested second copies of letters and information packs at this stage, while others declined to take part.

Contacting YP who had expressed an interest in the study involved using various methods such as email, phone, voicemail and text message depending on their preferred method of contact. This also required additional follow-up, and after a

few attempts contact was deemed unsuccessful for some who had given consent to be contacted. Generally though, when contact was successful YP were keen to participate and support the research. As half the sample were at university interviews had to be scheduled around exams and often took place during university holidays, or outside of working hours for those in employment. Three YP expressed an interest in participating but were unable to maintain the interview due to work commitments, geographical location and one person not feeling ready to talk about their experience due to other life events.

Despite the challenges of recruiting a sample of YP researchers should persist to ensure their voices continue to be heard and represented. In order for recruitment to be successful researchers should ensure the availability of resources to follow-up initial contact and allow time to be flexible with data collection to fit around the priorities and needs of participants.

Extended discussion

This section aims to bring together and elaborate the findings of the systematic review and empirical study, to provide a critique of each and to the consider the resulting implications for further research. Relevant clinical and theoretical implications are also discussed.

Thesis rationale

This thesis aimed to advance our understanding of YP's perspectives on end of pathway cleft surgery, thereby contributing knowledge to the field of craniofacial research. Both the systematic review and empirical study were undertaken in light of recognition that prevailing literature has tended to focus upon the clinician-reported outcomes of treatment (Eckstein et al., 2011; Stock et al., 2015). There has also been

limited empirical attention devoted to end of pathway cleft surgeries despite their occurrence at a developmentally sensitive period in the young person's life.

Furthermore the majority of the literature has neglected to obtain the views of YP transitioning to adulthood (Nelson, 2009). By reviewing the literature on the patient perspective and by employing a qualitative approach this thesis aimed to offer new insights into YP's experience of elective CL/P surgery at the end of the treatment pathway. Measuring outcomes and developing an understanding of patient experiences is vital to ensuring treatment effectiveness (Hens et al., 2011; Porter, 2010; Jenkinson, Coulter, & Bruster, 2002).

Summary of key findings

Overall the thesis highlights that YP appear to derive some benefit from undergoing elective surgery at the end of the cleft treatment pathway, and they want their voices to be heard and autonomy respected in the process of deciding whether to undergo such surgery. Key findings from each paper are outlined and elaborated in the context of the current literature.

Systematic review. The review illuminated how studies evaluating PROs of cleft surgery tend to utilise either generic measures not validated in a cleft population or bespoke tools. Generic tools validated in the general population lack the sensitivity to detect concerns relevant to the cleft population (Crerand et al., 2017; Eckstein et al., 2011; Ricketts et al., 2016) and bespoke tools undoubtedly lack reliability, validity and responsiveness (Eckstein et al., 2011). Together this means that we are currently unable to draw firm conclusions about the PROs of end of pathway cleft surgery. This reinforces the need for the development of standardized

and cleft-specific measures to enable clinicians to effectively measure the PROs of cleft surgery.

Furthermore, the variability in terms of what measures are used and at what time point adds to the difficulty in determining PROs. The studies reviewed indicate that patients derive some benefits from undergoing end of pathway surgery (e.g. improved satisfaction with appearance, quality of life and social interaction) however such conclusions are tentative due to concerns about the way outcomes were measured. Also highlighted by the review is the lack of guidance about the optimum post-surgery window when outcomes are best captured, evidenced by the inconsistency of when outcomes were measured across the studies. This makes it difficult to compare outcomes across studies and also limits understanding about when potential benefits of surgery emerge, and whether they are maintained or change over time. It also means clinical teams are likely to be measuring PROs ineffectively or perhaps not measuring them at all due to lack of appropriate tools and guidance about how and when to measure outcomes. For example, one study in this review measured satisfaction 12-weeks post-surgery and did not find significantly increased levels of satisfaction (Cheung et al., 2006). Whilst this could be representative of nominal variation in satisfaction pre and post-surgery it could also be due to patients needing time to recover from the effects of surgery and adjust to their altered appearance.

The methodological flaws evident in the majority of studies included in this review echo the findings of other related reviews (Hunt et al., 2005; Stock & Feragen, 2016) and are perhaps indicative of the challenges of carrying out high-quality research within this field (Chung & Burns, 2008). A recent paper brings into

sharper focus some of the conceptual and methodological difficulties that plague the field of craniofacial research (Stock et al., 2018), many of which are relevant to the findings of the present review. In particular, the authors suggest that ‘psychological adjustment’ as a transitory concept is hard to define and measure especially with the lack of consensus about what or where the adjustment ‘end point’ is.

Empirical study. Thematic analysis revealed that for YP in this study decision making was experienced as a multifaceted process. Contextual and personal factors such as perceived societal or authoritative pressure, the support of others, the nature of cleft treatment pathway, self-perception and identity help explain how YP experienced the process of deciding about OS. Mostly it was experienced as a big decision and one symbolic of developing a sense of adult maturity and autonomy. Although some welcomed the opportunity for surgery, for a few it was a difficult decision as it challenged their view of themselves as “normal” by drawing attention to difference. Indeed, it is suggested that definitive surgeries at the end of the cleft pathway aim to provide a level of invisibility from the cleft identity (Cadogan & Bennun, 2011), to rectify feelings of difference and to improve self-perception (Alansari et al., 2014). This is supported by the fact that all YP opted to have some additional surgery regardless of whether it was OS, however such an inference is tentative given that the motivations for undergoing OS were not explicitly explored.

Unfortunately, professionals use of language pertaining to “correction” served to further emphasise YP’s sense of difference. In the field of visible difference particularly it is advocated to use thoughtful and sensitive terminology so as not to further stigmatise affected individuals, hence use of the term ‘visible difference’ to replace negatively framed terms such as ‘deformity’ (Rumsey & Harcourt, 2007).

Sensitive use of language is crucial in a context where frequent and long-term contact with the clinical team often socialises patients to surgery as a method to improving quality of life (Kapp-Simon et al., 2015; Strauss et al., 2007). In the present study not all YP felt jaw surgery was truly optional in the same way they felt nose and lip surgery was, yet for many YP the jaw was not their primary feature of concern. Recent research suggested that although YP with CL/P were more concerned about aspects of facial appearance, they had developmentally normative appearance-related concerns and invest less in their overall appearance than non-affected peers (Crerand et al., 2017). This highlights the need for YP to be involved in the decision making process as they may have differing views, priorities or values to others.

Findings emphasize ways in which YP and their families can be effectively prepared for and supported through the decision making process. For example, the developmental involvement of children and young people in decisions about cleft treatment is recommended to help prepare YP to be competent decision makers. In addition, YP particularly valued the support of others when they did not impose their own opinions and instead reinforced the autonomy of the YP as decision maker and offered a space for YP to talk through and weigh up their decision. Findings also highlight the need for clinical teams to make the optional nature of OS more explicit by normalising the option of not having OS, and also making explicit that OS can be delayed. Doing so might work towards challenging the culture of similarity that operates in our society (Strauss et al., 2007). It may also lead to lower uptake of elective surgery if shared decision making was effectively employed (Boss et al., 2016).

Thesis strengths and limitations

To the authors knowledge both the systematic review and empirical study are the first to explore two neglected areas in the field, namely YP's perspectives and end of pathway cleft surgery. Specific strengths and limitations pertaining to each paper are further outlined below.

Systematic review. Due to known methodological challenges within the field broad inclusion criteria were set to ensure a comprehensive overview of the current literature (Stock & Feragen, 2016). This can be considered a key strength as this inclusive approach has led to an improved understanding of current practice and the quality of available evidence to help guide future research. The credibility of findings and quality of the review was improved by having a second reviewer independently check identified papers for inclusion and by performing an inter-rater quality check on a proportion of the included papers. To increase reliability a second researcher could have independently extracted the data from each included paper or checked the proforma for accuracy.

Unfortunately, due to time and resource limitations the findings reported are restricted to studies written or translated into the English language, thereby potentially excluding other relevant literature. Similarly, the grey literature was not searched so the review is limited by publication bias and important or contradictory findings may have been missed. It is suggested that unpublished literature can be equally as rigorous (Lipsey & Wilson, 2001), therefore future reviews might aim to include grey literature especially given the limited research in this field. A further limitation is that authors of included studies were not contacted; doing so may have

led to the inclusion of studies that were excluded because the cleft subsample data was not filtered.

Empirical study. Prior studies acknowledge a need for direct and in-depth exploration of YP's decision making experiences of additional cleft surgery (Alderson, 2006; Hall et al., 2012; Kapp-Simon et al., 2015). The use of a qualitative approach for the empirical study achieved this aim and enabled exploration of how YP found the process of making a decision, which helps to counter the criticism that much decision making research explores competence rather than the process around making a decision (Sugarman et al., 1999). In addition, conducting the analysis inductively meant themes were driven by the data rather than from existing theory, which helped to prioritise the patient perspective. It is acknowledged however, that the analysis and interpretation may be affected by the researcher having engaged with some of the literature before analysis; though some suggest this can be helpful in increasing sensitivity to alternative aspects of the data (Tuckett, 2005). Participants were verbally informed when providing consent that their accounts would be subject to interpretation and they were offered an opportunity to receive a summary of the key findings, which 11 out of 12 participants opted to receive. Disseminating findings and sharing them with participants is considered a key component of good research ethics.

Credibility checks for the thematic analysis were limited to discussions with the research supervisor and reflections from the clinical collaborator. One method for increasing credibility might have been to involve participants in reviewing the analysis and providing feedback (Lincoln & Guba, 1985). However, for the present study so called 'member checking' was not part of the research design and the

timescale of the research meant this would not have been possible. There is also much criticism about member checking as ontologically it aligns with the realist framework since participants are asked to agree with or refute the analysis as representative of their reality (Sandelowski, 1993; Tracy, 2010), thereby contradicting the interpretative nature of qualitative research. In addition, the validity of member checking has been questioned as participants are likely to be reluctant to provide honest feedback due to the power dynamic or equally, they may be less able to bracket off their own views meaning feedback provided would be biased by their own agendas (McLeod, 2001). 'Member reflections' are proposed as an alternative where members elaborate the findings rather than establishing their credibility (Tracy, 2010). This approach acknowledges that participants will see, read and understand things differently to the researcher. Using this approach may have given more depth to the analysis for the empirical study and will be borne in mind for future research.

It is noted that the ethnic diversity of the participants was limited and there were more males, although this appeared generally representative of the eligible sample. Finally, incorporating patient representatives into the research process helped improve the quality and validity of this research by ensuring the readability and appropriateness of research materials and ensuring the study's relevance to the population under study.

Theoretical implications

The theoretical implications arising from this thesis broadly relate to and support current understandings about the psychology of appearance, psychology of choice and models of decision making.

Psychology of appearance. Despite the known effects of visible differences on psychosocial wellbeing the psychology of appearance is a topic that has historically received limited attention in the literature (Rumsey, 2008). As such the experience of living with a cleft or other visible difference is not captured by a particular model (Norman & Moss, 2014). It is also observed that relatively few studies in the field apply psychological theory to their findings (Feragen & Stock, 2018). The findings within this thesis are perhaps best captured by aspects of the cognitive-behavioural model of body image development (Cash, 2012) which has recently been applied to the craniofacial field (Feragen & Stock, 2018). Cash's model aims to summarise the complex range of processes involved in determining an individual's satisfaction with their appearance or body image, including predisposing factors (socio-cultural and interpersonal experiences, personality and physical characteristics) and proximal factors (cognitive appraisals of appearance, self-regulatory strategies, activating events such as appearance-altering treatment).

In particular, the decision making experiences of young people, the way surgery was discussed and ultimately psychosocial outcomes and satisfaction with surgery appear to map onto aspects of the cognitive-behavioural model of body image development. Relevant aspects include those which highlight how the societal pressure to 'look good' and conform to the socially construed ideal of facial attractiveness affect people differently. This was evident in YP's experiences in the way that some pursued surgery to fit in, whereas others did not feel this pressure. The way in which the decision was discussed with parents and professionals highlights how it is not only patients who will be affected by socio-cultural ideals of facial appearance, and this was evidenced to some extent by the reported positive bias towards undergoing OS. It is also likely that the PROs and satisfaction with

surgery will depend upon patients' interpersonal experiences, personality, self / other appraisals and investment in appearance, although this was not explicitly explored in either the review or study, and needs further investigation (Feragen & Stock, 2018).

Finally, Cash's model proposes how appearance schemas can be triggered by a range of activating events, of which discussing and undergoing appearance altering surgery would be one. Findings suggested the activation of appearance schemas was likely for YP in the empirical study in the way that some began to doubt their appearance once the 'difference' was pointed out by the clinical team.

Psychology of choice and models of decision making. Broadly, findings from the empirical study support descriptive models of decision making which emphasise the use of heuristics (Chaiken, 1980). YP's narratives indicated the use of heuristics to simplify the decision and attend to certain aspect of information in line with their preferences and values. In addition, findings appear to uphold the theory that the framing of information affects how choices are perceived and made (Tversky & Kahneman, 1981); this was evidenced by YP's experience of OS being positively framed or it not being perceived as a genuine option. In addition, the finding that a number of patients opted for OS because they felt they would be 'missing out' by deciding not to have it is congruent with the regret theory of decision making (Loomes & Sugden, 1987). In line with regret theory YP appeared to consider not only the consequences of having the surgery but also the regret they might experience if they did not.

The way YP experienced the decision making process and the way they made decisions appeared to be shaped by predisposing factors (e.g. previous involvement in treatment), patient factors (personality and interest in the process), the attitude of parents and professionals to the decision and the context in which the decision was

made (e.g. the nature of cleft pathway), which aligns with ideas in the literature about decision making competence (Mårtenson & Fägerskiöld, 2008; Miller et al., 2004).

It was clear that most YP wanted to be involved in decisions, they wanted to matter to clinicians and ideally wanted to feel informed about aspects of the treatment that mattered to them. This corresponds with models of shared decision making (SDM) which highlight the need for professionals to value patients own expertise and explore their values and preferences (Barry et al., 2012; Coulter, 1999; Coulter & Collins, 2011). However, the extent to which cleft services are set up to support SDM may warrant direct investigation.

As current theoretical perspectives do not seem to adequately account for the multifaceted nature of the decision making process, further work could be done to develop a model of patient decision making which better depicts the complexity inherent in healthcare decisions, especially when the treatments on offer are not urgently necessary and based on patient choice (e.g. prior life and treatment experiences, feelings, thoughts, appraisals of appearance, contextual, social and identity factors).

Clinical implications

As this thesis focused upon a specific population transferring findings more broadly is difficult, however findings indicate the need to find more effective methods of measuring PROs and for YP to feel adequately informed and supported in their decision making for elective surgery. Such implications could be applied to other visible differences where elective surgeries are available.

Measuring PROs. To inform practice and research, clinical consensus is

needed and guidance developed about the key time-points to measure PROs following end of pathway cleft surgery. In addition, consensus is needed from the cleft community about which tools can most effectively measure PROs of surgery. The development of the CLEFT-Q (Tsangaris et al., 2017; Wong Riff et al., 2017) may well fulfil this need.

Once it is established how and when PROs are best measured, it is recommended that cleft teams, with the support of psychologist colleagues, routinely measure PROs (e.g. psychosocial adjustment and satisfaction with facial appearance) at key developmental transition points (Hearst et al., 2010) and prior to discussing elective surgery. Measuring PROs (e.g. psychosocial functioning and satisfaction with facial appearance) before discussions are held about OS (and other elective surgeries) would help inform teams about patients' self-perception and may reduce the chances of patients undergoing surgery because it is 'recommended'. It would also provide an opportunity to establish whether desire for surgery masks underlying concerns that could be best worked through psychologically before surgery is pursued (Alansari et al., 2014; Sinko et al., 2005). Such recommendations are not new – Marsh (1990) in considering when enough surgery is enough made similar recommendations 28 years previously.

As well as using a validated cleft-sensitive measure it is proposed that researchers may wish to also utilise a generic measure to enable comparison of outcomes with non-affected peers (Crerand et al., 2017) undergoing comparable surgery. Crerand et al. (2017) suggest that doing so would help to better contextualise any reported psychosocial difficulties in line with what may be considered normative among peers for that period of development.

Facilitating decision making. Findings from the empirical study reinforce the need for professionals and parents to developmentally involve children and YP in decisions about treatment across the pathway, but especially in the case of elective surgery. It is acknowledged however that patients may vary in the degree to which they want to participate and have control, so involvement is best determined by the patient's individual preferences (Guadagnoli & Ward, 1998). Involving patients as partners in their treatment from a young age and emphasising the genuinely elective nature of end of pathway cleft surgery is likely to help patients feel more empowered to say when they feel content with the face they have (Marsh, 1990; Strauss et al., 2007). It is possible that the use of decision aids may prove useful in order to help professionals present balanced information and to help patients assimilate information.

With psychologists as members of the cleft multi-disciplinary team they are well-placed to be able to provide a confidential space for YP to discuss, weigh-up and emotionally process the decision about whether to undergo further surgery (Stock et al., 2016). Where necessary, psychologists may also be able to offer support to parents where there is difficulty relinquishing responsibility (Hearst et al., 2010).

Moreover, careful and sensitive use of language is also recommended when discussing elective surgery to reduce the chance that patients perceive the mention of further surgery as intimating they need it. This is especially important given the power dynamics inherent in patient-doctor consultations and the nature of the cleft pathway. Psychologists and patient representatives may be able to support clinicians

in developing non-stigmatising language that would help rebalance the power and minimise difference in cleft consultations.

Research implications

Findings from both the review and study earmark areas for further research, some of which are aforementioned in each paper. Ultimately, to enable firmer conclusions to be drawn about the PROs of end of pathway cleft surgery the field would benefit from conducting well-designed, longitudinal studies.

As the empirical study only captured the decision making experiences of the patient, and because both parents and professionals are key to facilitating and supporting decision making, researchers would be encouraged to capture their experiences to provide a complete account of the process.

Quantitative studies using measures of self-concept and satisfaction with and investment in appearance would help to shed light on whether self-concept increases after elective cleft surgery, as was implied by a number of YP in this study. In addition, studies may wish to explore whether there are differences in self-perception between those who choose to have OS and those who decide not to have OS. Studies aimed at determining the factors responsible for positive self-perception will be particularly important in an era where medical advances are ongoing, and where there is a possibility of continual revision surgeries.

Overall conclusions

Taken as a whole this research provides a much needed insight into the experiences of YP deciding about a definitive elective surgery in the context of the CL/P treatment pathway, as well as an overview of the current literature reporting the PROs of end of pathway cleft surgery.

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List of appendices

Appendix 1. The Cleft Palate-Craniofacial Journal Manuscript Preparation

Guidelines

Appendix 2. Data extraction template

Appendix 3. Quality criteria for critical appraisal of included studies

Appendix 4. Quality criteria rating table with inter-rater checks

Appendix 5. Table of excluded studies with reasons

Appendix 6. Health Research Authority ethical approval

Appendix 7. Interview guide

Appendix 8. Patient invitation letter

Appendix 9. Recruitment advert

Appendix 10. Participant information sheet

Appendix 11. Participant consent form

Appendix 12. Demographic information form

Appendix 13. Example of coded transcript extract

Appendix 14. Example coding and theme development

Appendix 15. Supplementary participant quotes to support derived themes

Appendix 1. The Cleft Palate-Craniofacial Journal Manuscript Preparation Guidelines



The Cleft Palate-Craniofacial Journal

MANUSCRIPT PREPARATION

GENERAL INFORMATION

SCOPE

The Cleft Palate-Craniofacial Journal (CPCJ) is directed to a multidisciplinary readership of clinicians and scientists interested in craniofacial anomalies, including cleft lip and cleft palate. The CPCJ publishes original research articles, clinical reports, brief communications, articles related to new ideas or innovations, letters to the editor, editorials, invited book reviews, and meeting announcements.

CONTACT INFORMATION

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Submission of a manuscript to the CPCJ is taken as evidence that no portion of the text or figures has been published or submitted for publication elsewhere unless information regarding previous publication is explicitly cited and written copyright permission obtained and uploaded at the time of manuscript submission. Permission should be obtained for both print and online publication.

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Two independent peer reviews are typically solicited. At the discretion of the Section Editor, a third 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 and with high levels of evidence may be advanced in the publication queue

at the discretion of the Editor.

All submitted articles are “double-blinded” 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.

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Publication of material in the CPCJ should not be interpreted as an endorsement of the material contained therein. The publisher will send galley proofs of accepted manuscripts (pdf file) to the corresponding author via email. Corrections and revisions should be returned to the publisher via email as instructed. Authors are responsible for the accuracy of references and statistical computations. The author(s) acknowledge that the publisher reserves the right to charge authors for excessive revisions made to their galley proofs. Publication may be withheld if authors fail to fulfill these financial obligations.

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The corresponding author is responsible for ensuring that all individuals named as co-authors have made a major contribution to the manuscript. Authorship credit should be based on significant contributions to 1) conception and design, or acquisition of data, or analysis and interpretation of data, AND 2) either drafting of the manuscript or critical revision of the manuscript for important intellectual content, AND 3) final approval of submitted manuscript. Each author must declare his or her contribution to the manuscript by signing the copyright transfer form.

FINANCIAL DISCLOSURE/CONFLICT OF INTEREST

Authors are required to disclose, in a cover letter accompanying their manuscript, 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 at the discretion of the Editor, may be included in publication of an accepted manuscript.

GROUP AUTHORSHIP

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 in the Author Responsibility section above. Other research group members who do not qualify for authorship may be listed in an Acknowledgement.

PATIENT ANONYMITY

The author is responsible for ensuring the anonymity protection of any individual depicted in a manuscript. A signed permission form must be submitted for any recognizable individual appearing in manuscript figures. Shading of the eyes is not an acceptable means of rendering an individual unrecognizable. Patient permission forms may be downloaded from the CPCJ website (www.cpcjournal.org; see ‘For Authors’). 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. Consent forms should be uploaded as an 'Additional Information' file at submission.

HUMANS AND ANIMALS IN RESEARCH

For manuscripts describing the results of experimental studies on humans, authors must include a statement in the Methods section of the manuscript that a Human Subjects or Institutional Review Board (IRB) approved the study and that informed consent was obtained. 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 a formal IRB 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. Information regarding the Declaration of Helsinki may be found at <http://www.wma.net/en/30publications/10policies/b3/>.

All research involving animals must follow published guidelines for use of laboratory animals (<http://www.apa.org/science/leadership/care/guidelines.aspx>). Compliance with these guidelines should be indicated in the Methods section of the manuscript, along with Institutional Review Board approval if appropriate.

MANUSCRIPT PREPARATION

SUBMISSION CATEGORIES

Original Articles: 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. Limit: 7 typeset pages as they appear in the journal (about 7,000 manuscript words, with up to 6 figures or tables combined).

NEW IN 2016: *What I (We) Do:* Introduce new solutions to clinical problems. Novelty and quality of illustrations and videos (when appropriate) are key ingredients. Authors should include a brief (50-75 words) abstract with the following format: Background (what is the issue/problem), solution, what I/we did that is new. Also include 3-5 key words. If no patient identifiable data are included, no IRB form is necessary. Limit: 2 typeset pages as they appear in the journal (about 1,000 words, with up to 3 figures or tables combined, and up to 5 references).

Clinical Reports: Case reports presenting new clinical information. Limit: 4 typeset pages as they appear in the journal (about 4,000 manuscript words, with up to 6 figures or tables combined).

Ideas and Innovations: Short communications related to novel ideas, techniques, methods of assessment, etc. Limit: 3 typeset pages as they appear in the journal (about 3,000 manuscript words, with up to 3 figures or tables combined).

Brief Communications: Preliminary or limited results of original research pertaining to prevalence, causes, mechanisms, diagnosis, course, treatment, and prevention. Limit: 3 typeset pages as they appear in the journal (about 3,000 manuscript words, with up to 3 figures or tables 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. Limit: 3 typeset pages as they appear in the journal (about 3,000 manuscript words, with up to 3 figures or tables combined).

Perspectives are typically solicited articles (unsolicited articles will be considered) that provide background and context for an article in the issue in which they appear. Limit: 1.5 typeset pages as they appear in the journal (about 1,500 manuscript words, with up to 1 figure or table).

Letters to the Editor: Comments in the form of letters that express differences of opinion or supporting views of recently published CPCJ content. Limit: 1.5 typeset pages as they appear in the journal (about 1,500 manuscript words, with up to 1 figure or table).

Editorials: Brief substantiated commentaries on subjects of interest to the CPCJ readership. Editorials should be narrative in form. Limit: 1.5 typeset pages as they appear in the journal (about 1,500 manuscript words, with up to 1 figure or table).

MANUSCRIPT FILES TO BE UPLOADED**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
- Academic rank or position, and institutional affiliation for each author
- Name, address, telephone number, fax number, and email address of the corresponding author, who will receive all editorial communication and reprint requests.
- If applicable, statement that manuscript was presented orally at a professional meeting, including the name, date, and location of the meeting.
- Credits and appropriate grant numbers if the study was supported by an agency.
- Running title (less than 8 words).
- If applicable, statement acknowledging all forms of financial support
- If desired, any other acknowledgements (e.g. individuals assisting with conduct of the study but not qualifying for authorship).

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

2. Manuscript**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 (including Key Words) with the following headings and information, as applicable. Structured abstracts of no longer than 150 words should be used for data-based Brief Communications articles.

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, blinding, 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 blinding. 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).

Key Words: A short list of the key words that reflects the article’s content.

Clinical reports should include an unstructured abstract of no longer than 100 words, including Key Words, describing the objective, essential features and uniqueness of the case being presented, and conclusions. Non-data-based Brief Communications and Ethics, Legal, or Health Policy reports should include an unstructured abstract of no longer than 100 words, including Key Words.

Page 3:

Where applicable, divide the body of the manuscript into the Introduction, Methods, Results, Conclusion, and 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, beginning with the second page. 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.

<u>ABBREVIATION</u>	<u>USED TO DESCRIBE A SUBJECT GROUP THAT INCLUDES:</u>
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)
CP±A	cleft palate with or without cleft alveolus (excludes (1) cleft lip, (2) cleft lip and alveolus, and (3) cleft lip and palate)

TERMS THAT MAY BE ADDED TO THE ABBREVIATIONS ABOVE (IF APPROPRIATE):

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/ReferencesSingle Author Article

Citation: Mantel (1963) or (Mantel, 1963)

Reference: Mantel N. Chi-square tests with one degree of freedom; extensions of the Mantel-Haenszel procedure. *J Am Stat Assoc.* 1963;58:690–700.

Two Author Article

Citation: Rasheed and Munshi (1996) or (Rasheed and Munshi, 1996)

Reference: Rasheed SA, Munshi AK. Electromyographic and ultrasonographic evaluation of the circum-oral musculature in children. *J Clin Pediatr Dent.* 1996;20:305-311.

Three Or More Author Article

Citation: Lilja et al. (2000) or (Lilja et al., 2000)

Reference: Lilja J, Elander A, Lohmander A, Persson C. Isolated cleft palate and submucous cleft palate. *Oral Maxillofac Surg Clin N Am.* 2000;12:455–468.

Two or more works by the same first author in the same year

Citation: Smith (1975a), Smith (1975b) or (Smith, 1975a) etc

Reference: Smith RC. Long term effects of smoking on fetal development. *Teratology* 1975a;42:75-84.

Monograph

Citation: Bardach (1967) or (Bardach, 1967)

Reference: Bardach J. *Cleft Lip and Palate* (Monograph). Warsaw: Polish Institute of Medical Publications; 1967.

Thesis

Citation: Dowden (1992)

Reference: Dowden PA. The Effects of Listener Training on the Speech Intelligibility of Severely Dysarthric Individuals. Seattle, WA: University of Washington; 1992. Dissertation.

Book

Citation: McWilliams et al. (1990) or (McWilliams et al., 1990)

Reference: McWilliams BJ, Morris HL, Shelton RL. *Cleft Palate Speech*. Philadelphia: BC Decker; 1990: 40-49. (only list pages if specific pages are cited).

Chapter in Book

Citation: Eliason (1990) or (Eliason, 1990)

Reference: Eliason MJ. Neuropsychological perspectives of cleft lip and palate. In: Bardach J, Morris HL, eds. *Multidisciplinary Management of Cleft Lip and Palate*. Philadelphia: WB Saunders; 1990:825–831.

Conference Presentation

Citation: Parke and Sawin (1975) or (Parke and Sawin, 1975)

Reference: Parke RD, Sawin DB. Infant characteristics and behavior as elicitors of maternal and paternal responsivity in the newborn period. Presented at the Meeting of the Society for Research in Child Development; April 1975; Denver, Colorado.

Website

Citation: World Health Organization (2005)

Reference: World Health Organization. International database on craniofacial anomalies. Available at: www.who.int/genomics/anomalies/. Accessed June 27, 2005.

When multiple references are cited simultaneously in the text, they should be arranged in chronological order, for example: (Smith, 1975; Jones et al., 1981; Brown, 1986). References should be double-spaced, and listed in alphabetical order (unnumbered) according to the surname of the first author. For articles with more than ten authors, include only the first ten author names in the reference list, followed by “et al.”.

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.

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.

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 upload that permission as an “Additional Information” file at submission. Figures should be numbered consecutively in the order in which they appear in the manuscript, using Arabic numerals. A “List of Figure” Legends must be included on a separate page following the body of the manuscript. The legend should explain each figure in detail. Authors will be responsible for the following charges for each color figure submitted: \$75.00 for online only; \$400.00 for both online and print for ACPA members or \$500.00 for non-members. A single figure may include multiple images (a, b, c, etc.) but all must appear on the same page.

Figures should be submitted in one of the following formats: tif (preferable), eps, jpg, pdf. Each figure should be submitted as a separate file. Composite figures made up of more than one image should be submitted as separate files (e.g. Fig 1A, Fig 1B). However, composite figures should contain a single legend describing the contents of all figures in the composite.

Refer to the Digital Art Specifications document at www.cpcjournal.org (see ‘For Authors’) for image resolution, size, and format requirements. For symbols that must be explained, please use a key that can be shot with the figures. Do not include symbols in the figure legend. Authors may be charged if artwork must be generated to incorporate figure symbols into the figure legend.

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.

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 6 MB in

size. If submitting avi files, the files must be compressed. Authors are solely responsible for all editing of video clips. 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.*

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 length. 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.

Appendix 3. Quality criteria for critical appraisal of included studies

Criteria	Definition	Rating (2 = Well-addressed 1 = Partially addressed 0 = Poorly addressed/ Not addressed/Not reported)
1. Study aims	Study aims are easily identified and clearly described in introduction / method	2
	Study aims are vaguely described not easily apparent from the introduction and method	1
	Study aims unclear / not reported	0
2. Study design	Study design evident and appropriate to address aims of study	2
	Study design not clearly identified (but not inappropriate) / study design only partially addresses study aims	1
	Design inappropriate to address study aims	0
3. Sample Size	Sample size seems appropriate to design and outcome under study. Where appropriate, power and effect sizes reported.	2
	Sample size appears small and no mention of power/effect size despite reporting some statistically significant results	1
	Sample size is obviously inadequate to draw more generalized conclusions	0
4. Sample characteristics	Selected sample relevant with defined inclusion criteria and clearly described demographic information	2
	Selected sample relevant, selection methods may be unclear / some demographic information not collected/not clearly reported	1
	Selected sample poorly described (e.g. study could not be replicated)	0
	Surgical intervention/s clearly described.	2

5. Surgical intervention/s	Surgical intervention/s described, however specific details not provided or some aspects unclear.	1
	Surgical intervention/s not clearly described, or no description provided.	0
6. Patient-reported outcome measure/s used	Use of standardised outcome measure/s validated in cleft population	2
	Use of standardised outcome measure/s, not validated in cleft population	1
	Use of non-standardised bespoke measure (e.g. basic satisfaction scale)	0
7. Validity of when outcomes measured	Measures administered pre and post-surgery (≥ 6 months post-surgery) and after appropriate follow-up period (6 months – 2+ years post-surgery)	2
	Measures administered pre and post-surgery only (≥ 6 months post-surgery) / measures administered pre and post and after inappropriate follow-up (< 6 months post-surgery)	1
	Measures administered retrospectively or at only one time point	0
8. Analysis	Analysis appropriate to the study design and type of outcome measure used. If appropriate, missing data handled appropriately and effect sizes reported	2
	Only descriptive statistics used due to study design OR analysis not clearly described but likely appropriate (e.g. parametric tests used, but unsure from details provided if appropriate. If relevant, no or unclear management of missing data and effect sizes not reported	1
	Analysis not clearly described, or not obviously appropriate	0
9. Conclusions	Conclusions drawn are supported by the study results, and appropriate caution is used in conclusions made.	2

	Some of the conclusions are supported by the data, others are not. Caution may not be evident in drawing conclusions from the results (e.g. given sample size and generalizability)	1
	Few or none of the conclusions are supported by the data.	0

*based on the SIGN-50 quality rating tool

Overall quality ranking

≥ 13	++ Good	All or most criteria fulfilled, and if not, then conclusions unlikely to alter.
9-12	+ Acceptable	Some criteria fulfilled but some only partially fulfilled, or some criteria not met at all. Limitations may modestly affect the findings and conclusions.
≤ 8	- Poor	Few criteria fulfilled, concerns about methodological quality and ability to draw conclusions about psychosocial outcomes.

Appendix 4. Quality criteria rating table with inter-rater checks

Criteria→ (inter-rater score)	1.Study aims	2.Study design	3.Sample Size	4. Sample characteristics	5. Surgical intervention/s	6.Psychosocial outcome measure used	7.Validity of when outcomes measured	8.Analysis	9.Conclusions	Total score
Study										
Albers et al. (2016)	2	1	1	2	0	1	0	1	1	9
Karabekmez et al. (2015)	2 (2)	1 (1)	0 (0)	2 (2)	2 (2)	0 (0)	0 (0)	1 (1)	2 (2)	10 (10)
Gassling et al. (2015)	2	2	0	2	2	1	0	2	1	12
Byrne et al. (2014)	2	2	1	2	2	1	0	1	2	13
Roosenboom et al. (2014)	2	2	1	2	2	1	1	1	2	14
Chua et al. (2012)	2	2	2	2	0	1	2	1	2	14
Hens et al. (2011)	2 (2)	2 (2)	1 (1)	2 (2)	2 (2)	1 (1)	0 (0)	2 (2)	2 (2)	14 (14)
Eggermont et al. (2007)	2	1	0	2	2	0	0	1	2	10
Cheung et al. (2006)	2	2	0	2	0	1	1	1	2	11
Andersen et al. (2012)	2	2	0	2	2	0	0	1	2	11
Balaji (2016)	1	2	1	2	2	1	0	1	1	11
Ricketts et al. (2016)	2 (2)	2 (2)	1 (1)	2 (2)	0 (0)	1 (1)	1 (1)	2 (2)	2 (2)	13 (13)
Vass et al. (2016)	2	2	0	2	2	0	0	1	1	10

Pausch et al. (2016)	2	2	2	2	2	0	0	1	2	13
Sawyer et al. (2016)	2	2	1	2	2	1	1	1	2	14
Pitak-Arnnpot et al. (2011)	2	2	1	2	2	0	0	2	2	13
Tiong et al. (2014)	1 (1)	1(1)	0 (0)	2 (2)	2 (2)	0 (0)	1(1)	1(1)	1(1)	9 (9)
Chaithanyaa et al. (2011)	2	2	0	2	2	0	0	1	1	10
Sandor et al. (2006)	2	2	1	2	1	0	0	1	2	11
Jones et al. (2017)	2	2	0	2	2	0	0	0	1	9
Scopelliti et al. (2013)	0 (0)	1(1)	1(1)	1(1)	0 (0)	0 (0)	0 (0)	0 (0)	1 (1)	4 (4)
Simon et al. (2016)	1	1	0	1	2	0	0	1	1	7

Appendix 5. Table of excluded studies with reasons

Study	Reason for exclusion
Antoun, Fowler, Jack, & Farella (2015).	The intervention for the CL/P subsample was orthodontic treatment not surgery.
Germec-Cakan, Canter, Cakan, & Demir (2014).	Does not include a pre or post-surgery patient-reported outcome measure, authors just state patient was satisfied.
Stork, Kim, Regennitter, & Keller (2013).	Outcome data on cleft subsample is not filtered so cannot determine the results for the cleft population.
Mokal & Juneja (2014).	Does not include a pre or post-surgery patient-reported outcome measure, authors just state patients were satisfied.
Bhuskute et al. (2017)	Majority of surgery took place with children (mean age =7.9 years; range 4-20 years), so not end of pathway and outcomes not split by age.
Oosterkamp et al. (2007)	Considers general treatment outcomes rather than surgery specific outcomes and does not evidently concern end of pathway.
Munz, Edwards, & Inglehart (2011).	Considers general treatment outcomes rather than surgery specific outcomes.
Lucchese, Gherlone, Asperio, & Baena (2014).	Does not include a pre or post-surgery patient-reported outcome measure.
Jeong, Lee, & Shin (2012).	Does not include a pre or post-surgery patient-reported outcome measure.
Larsson, Becker, & Svensson (2013).	Not all participants were undergoing end of pathway surgery (mean age = 13.5 years; range = 6-22 years) and outcomes not clearly split by age.
Impieri et al. (2017)	Not all participants were undergoing end of pathway surgery (mean age = 19 years; range = 6-56 years) and outcomes are not clearly split by age and cleft subsample.

Appendix 6. Health Research Authority (HRA) ethical approval letter**Health Research Authority**

Miss Michelle Acum
10A The Brook
Sutton
Ely
CB6 2PU

Email: hra.approval@nhs.net

19 May 2017

Dear Miss Acum

Letter of HRA Approval

Study title: Young people's (YP) experience of the decision making process for orthognathic surgery in cleft lip and palate (CLP).
IRAS project ID: 211335
REC reference: 17/LO/0817
Sponsor: University of East Anglia

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

IRAS project ID	211335
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It is critical that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from www.hra.nhs.uk/hra-approval.

Appendices

The HRA Approval letter contains the following appendices:

- A – List of documents reviewed during HRA assessment
- B – Summary of HRA assessment

After HRA Approval

The document “*After Ethical Review – guidance for sponsors and investigators*”, issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, 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.

In addition to the guidance in the above, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the [HRA website](http://www.hra.nhs.uk), and emailed to hra.amendments@nhs.net.
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the [HRA website](http://www.hra.nhs.uk).

Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at <http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/>.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application

IRAS project ID	211335
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procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>.

HRA Training

We are pleased to welcome researchers and research management staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

Your IRAS project ID is 211335. Please quote this on all correspondence.

Yours sincerely,

Steph Blacklock
Senior Assessor

Email: hra.approval@nhs.net

Copy to: *Ms Tracy Moulton*

IRAS project ID	211335
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Appendix A - List of Documents

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

<i>Document</i>	<i>Version</i>	<i>Date</i>
Copies of advertisement materials for research participants [Participant Recruitment advert]	April2017	10 April 2017
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [UEA I&I letter_April2017]	v1_April2017	20 April 2017
GP/consultant information sheets or letters [GP notification letter_North Thames]	April2017	10 April 2017
GP/consultant information sheets or letters [GP notification letter_East]	April2017	10 April 2017
Interview schedules or topic guides for participants [Interview topic guide]	April2017	10 April 2017
IRAS Application Form [IRAS_Form_27042017]		27 April 2017
Letters of invitation to participant [Clinic recruitment letter_North Thames]	May2017v2	17 May 2017
Letters of invitation to participant [Clinic recruitment letter_East]	May2017v2	17 May 2017
Other [Demographic information sheet]	April2017	10 April 2017
Other [Statement of Activities]	1	17 May 2017
Other [Schedule of Events]	1	17 May 2017
Participant consent form [Consent form]	April2017	10 April 2017
Participant information sheet (PIS) [PIS_April2017]	April2017	10 April 2017
Participant information sheet (PIS) [PIS_May2017_v2]	May 2017_v2	17 May 2017
Referee's report or other scientific critique report [UEA proposal feedback]	July2016	20 June 2016
Research protocol or project proposal [Research Proposal]	April2017	10 April 2017
Summary CV for Chief Investigator (CI) [CV for chief investigator]	Nov2016	18 November 2016
Summary CV for student [CV_MAcum]	Nov2016	18 November 2016
Summary CV for supervisor (student research) [CV_JYoung]	Nov2016	11 November 2016
Summary CV for supervisor (student research) [CV_KMastro]	Nov2016	01 November 2016
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Lay summary]	April2017	10 April 2017

IRAS project ID	211335
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Appendix B - Summary of HRA Assessment

This appendix provides assurance to you, the sponsor and the NHS in England that the study, as reviewed for HRA Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England to assist in assessing and arranging capacity and capability.

For information on how the sponsor should be working with participating NHS organisations in England, please refer to the, *participating NHS organisations, capacity and capability and Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria) sections in this appendix.*

The following person is the sponsor contact for the purpose of addressing participating organisation questions relating to the study:

Name: Miss Michelle Acum

Email: m.acum@uea.ac.uk

HRA assessment criteria

Section	HRA Assessment Criteria	Compliant with Standards	Comments
1.1	IRAS application completed correctly	Yes	Applicant has clarified that the study type chosen is the correct type for this study.
2.1	Participant information/consent documents and consent process	Yes	No comments
3.1	Protocol assessment	Yes	No comments
4.1	Allocation of responsibilities and rights are agreed and documented	Yes	A Statement of Activities and Schedule of Events for use with the PIC sites has been provided by sponsor for use with the participating NHS organisations.
4.2	Insurance/indemnity arrangements assessed	Yes	Where applicable, independent contractors (e.g. General Practitioners) should ensure that the professional indemnity provided by their medical defence organisation covers the activities expected of them for this

IRAS project ID	211335
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Section	HRA Assessment Criteria	Compliant with Standards	Comments
			research study
4.3	Financial arrangements assessed	Yes	There is no external funding acquired for this study.
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	Applicant has clarified that updates to the participant information sheet will be made prior to study start.
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics Committee favourable opinion received for applicable studies	Yes	No comments
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England

<i>This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.</i>
<p>This is a qualitative methods only, student study with one NHS site type; PIC.</p> <p>The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. For NIHR CRN Portfolio studies, the Local LCRN contact should also be copied into this correspondence. For further guidance on working with participating NHS organisations please see the HRA website.</p> <p>If Chief Investigators, sponsors or Principal Investigators are asked to complete site level forms for</p>

IRAS project ID	211335
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participating NHS organisations in England which are not provided in IRAS or on the HRA website, the Chief Investigator, sponsor or Principal Investigator should notify the HRA immediately at hra.approval@nhs.net. The HRA will work with these organisations to achieve a consistent approach to information provision.

Confirmation of Capacity and Capability

This describes whether formal confirmation of capacity and capability is expected from participating NHS organisations in England.

Participating NHS organisations in England will be expected to formally confirm their capacity and capability to host this research.

- Following issue of this letter, participating NHS organisations in England may now confirm to the sponsor their capacity and capability to host this research, when ready to do so. How capacity and capability will be confirmed is detailed in the *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* section of this appendix.
- The [Assessing, Arranging, and Confirming](#) document on the HRA website provides further information for the sponsor and NHS organisations on assessing, arranging and confirming capacity and capability.

Principal Investigator Suitability

This confirms whether the sponsor position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and the minimum expectations for education, training and experience that PIs should meet (where applicable).

The Statement of Activities provided states that a local collaborator will be in place at the participating NHS organisations.

GCP training is not a generic training expectation, in line with the [HRA statement on training expectations](#).

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken

It is unlikely that letters of access or honorary research contracts will be applicable, except where local network staff employed by another Trust (or University) are involved (and then it is likely that arrangements are already in place). Where arrangements are not already in place, university staff (or similar) undertaking identification of potential participants would be expected to obtain an honorary research contract. This would 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.

IRAS project ID	211335
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Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England to aid study set-up.

- The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.

Appendix 7. Interview guide**INTERVIEW TOPIC GUIDE****Researcher introduction**

My name is Michelle Acum and I am a Trainee Clinical Psychologist at the University of East Anglia. I am carrying out a research project about how young people born with a cleft lip and or palate (CLP) experienced the process of deciding whether or not to have corrective jaw (orthognathic) surgery.

Thank you for agreeing to take part in this research. You have been invited to take part because you have valuable expertise about what it is like for a young person to make a decision about whether or not to have corrective jaw surgery. Young people's involvement in, and experience of, the decision making process for corrective jaw surgery is not something we know much about. By sharing your personal experiences, you will be helping us to understand what the process is like for young people and how we can potentially make it better for other young people who are offered corrective jaw surgery.

It is completely your choice whether you want to take part and share your experiences. You can decide at any point that you no longer wish to take part (without providing a reason) and this will not affect your care. It will be recorded on your hospital file that you have taken part in this research and met with me, however the information and answers you provide will be kept private, unless there are concerns about yours or someone else's safety. If so, we may need to speak to the cleft clinical team, but this would be discussed with you first.

You do not have to answer any questions you don't want to and please feel free to ask me at any time if something doesn't make sense.

Do you have any questions before we begin?

'Settling-in' questions

- Some young people (YP) find it helpful for me to know a bit about you as a person as well as talking about having a cleft. What would you like me to know about you that is important? It is up to you how much you share.
- People may refer to having been born with a cleft lip and or palate in different ways. What would you normally say?

RQ1: How did young people with a CLP experience the process of decision making for Orthognathic Surgery?***Corrective jaw (Orthognathic) surgery***

- As we're going to be talking about corrective jaw (orthognathic) surgery, it would be helpful to know what your understanding is. How would you explain what corrective jaw surgery in cleft lip and palate is to someone who does not know anything about it?

→ definition to ensure we share the same understanding.

Most people's jaws sit with the top over bottom, however people born with a cleft may have an underdeveloped top jaw meaning their top jaw sits back from their bottom jaw. People who have this jaw positioning are offered surgery to correct the positioning of their jaws so they are more balanced and their teeth bite together properly.

This surgery is elective as the person decides whether they wish to have the surgery or not. The surgery doesn't usually take place until people are around 16 years of age or older when most of the jaw growth is complete.]

- How did you first come to hear about corrective jaw surgery? *(who mentioned it? how did they explain it? was the optional nature of it clear to you?)*
- What do you remember about when corrective jaw surgery was first mentioned as a surgery option for you? *(How old were you? Who were you with? How did you feel? What went through your mind? Did you have lots of questions? How were these answered?)*

The decision making process

- What happened after corrective jaw surgery was first mentioned to you? *(further appointments? information? discussions?)*
- What steps were involved in the decision making process? *(what information or guidance were you given about making a decision?)*
- How would you describe your role as a young person in the decision making process? *(How much were you involved? Were your views taken seriously? How was this? How did you feel?)*
- Who else was involved in the process of reaching a decision? *(e.g. parents, siblings, other family, friends, cleft staff – surgeon, orthodontist, nurse, SALT, psychologist, other support agencies e.g. CLAPA)*
- Were your views similar or different to other people (e.g. family, clinical team?) *(How was this? How was it managed, by you and by others?)*
- At the time, how ready did you feel to make a decision about whether to have corrective jaw surgery? *(had you been involved in making treatment decisions before?)*

- How ready were other people for you to be involved in making decisions?
- How was a decision reached? (*key influences? what was important? What was difficult? Did anyone lead the decision making? How was this? How did this feel?*)
- Did you feel able to change your mind?
- As I'm really interested to understand your experience of making a decision – is there anything else about your experience that would be important for us to talk about?

RQ2: How can young people with a CLP and their families be best prepared for, and supported with, making decisions about Orthognathic Surgery?

- From your experience, was there anything that you found particularly helpful in preparing you to make a decision? (*had you expected to be making a decision? Did the clinical team or your parents do anything to prepare you?*)
- Could anything have been done differently to have prepared you for making a decision? (*Cleft team, family, what difference might this have made, what would this have meant to you?*)
- From your experience, was there anything that you found particularly helpful in how you were supported to make a decision? (*E.g. by your parents, family, friends, clinical team?*)
- Could anything have been done differently to support you in making a decision? (*Cleft team, family, what difference might this have made, what would this have meant to you?*)
- What are your thoughts on involving young people in decisions about corrective jaw surgery?

Reflections and feedback

- Is there anything else that would be important to talk about that we haven't covered?
- How have you found being interviewed and talking about your experiences?

Debrief

- Do you have any concerns about anything we have talked about today?
- Do you have any questions?

Appendix 8. Patient invitation letter⁹

[Hospital/cleft team headers and logos]
[Address]

[Patient name]
[Address]

[Date]

Dear [Name],

RE: Information about taking part in a research study looking at young people's experiences of the decision making process for corrective jaw (orthognathic) surgery.

I am writing to tell you about a study being conducted by Michelle Acum who is a Trainee Clinical Psychologist at the University of East Anglia. She is carrying out the study as part of her Doctorate in Clinical Psychology.

My colleague, Michelle, is interested in learning more about young people's experiences of deciding whether or not to have corrective jaw (orthognathic) surgery. I am therefore contacting you to let you know about the research in case you are interested in learning more. Enclosed are further details about the study.

If you are interested in learning more about this study, please read the enclosed information from Michelle, complete the enclosed reply slip and send it back to her in the pre-paid envelope. She will then contact you to talk to you about the research to see whether you would like to take part. You can also contact her by email, m.acum@uea.ac.uk or phone, [REDACTED] to express your interest.

By contacting her to express your interest you would be under no obligation to take part in the research. Taking part in the research is entirely voluntary. Also, it is important for you to know that whether you decide to take part or not would have no effect on the care and treatment you receive from your cleft team.

In a few weeks time, someone from the cleft team may contact you to see whether you have received the information. Thank you for considering this research invitation.

Yours sincerely,

Dr Sara O'Curry
Consultant Clinical Psychologist
Assistant Head, Paediatric Clinical Psychology and Counselling
Enc: Advert, Participant Information Sheet, Reply Slip.

⁹ May 2017_v2

Appropriate headers and logos

REPLY SLIP

‘Deciding about corrective jaw (orthognathic) surgery’

I am interested in learning more about the ‘Deciding about corrective jaw (orthognathic) surgery’ study and I agree that I am happy for Michelle Acum, the researcher, to contact me about this study.

Name:

Home telephone number:

Mobile telephone number:

Email address:

Address:

Best time to contact:

Please detach and return this slip to the researcher using the pre-paid envelope provided or pass it to a member of the cleft team. Alternatively, you can email or phone the researcher directly to express your interest in the study.

Appendix 9. Recruitment advert

WERE YOU BORN WITH A CLEFT LIP/PALATE?

Research participants needed...



to share their experiences of deciding whether or not to have corrective jaw (orthognathic) surgery.

This research project has received Health Research Authority (HRA) approval and favourable opinion from the London – Brent Research Ethics Committee (REC).
REC reference: 17/LO/0817
IRAS project ID: 211335

Interested to find out more?*

Please contact:

Michelle Acum
(Researcher) Email: [REDACTED]
Phone: [REDACTED]

OR:

[REDACTED]
(CLAPA) Email : [REDACTED]
@clapa.com
Phone: [REDACTED]

We would love to hear from you!

*By contacting us to find out more you will be under no obligation to take part in the study.

- 👤 **Who?**
 - ✓ You must be: a young person (aged 16-25 years) who is a patient of either the North Thames or East of England Cleft Service and have undergone or decided against corrective jaw (orthognathic) surgery in the last 6 months to 3 years.
- 🗨️ **What?**
 - ✓ Taking part in the research will involve a one-off, confidential interview where you will be asked questions about your experience of the decision making process for jaw surgery.
- 📍 **Where?**
 - ✓ The interview will take place at your local cleft clinic, at your home or, if preferred, over the telephone, at a time that suits you.
 - ✓ Unfortunately, we cannot pay travel expenses but as a thank you for giving up your time you will receive a £10 voucher.
- 🕒 **How long will it take?**
 - ✓ Around 60-90 minutes
- 🙋 **Why take part?**
 - ✓ By sharing your experiences you will be able to help services ensure the decision making process is as smooth as possible for other young people deciding about jaw surgery.

👤 **About the researcher**

- ✓ Michelle Acum is a Trainee Clinical Psychologist at the University of East Anglia. She is working with clinicians at East of England Cleft service and the North Thames Cleft service.



**University of
East Anglia**

Appendix 10. Participant information sheet¹⁰**PARTICIPANT INFORMATION SHEET****‘Deciding about corrective jaw (orthognathic) surgery’**

My name is Michelle Acum and I am a Trainee Clinical Psychologist at the University of East Anglia. I am carrying out a research project about how young people who were born with a cleft lip and or palate (CLP) experienced the process of deciding whether or not to have corrective jaw (orthognathic) surgery. This research project is being carried out as part of my Doctorate in Clinical Psychology. Please take time to read through this information carefully and ask me if anything is not clear or if you would like further information.

Why is it important?

We believe that you should be involved in treatment decisions about you that directly affect you. It is important for us to listen to and understand your experiences of deciding about corrective jaw (orthognathic) surgery, because it may help us to improve the way young people are prepared for and supported to make treatment decisions – this might mean changing things for the better or keeping them the same.

I would like to invite you to participate in this research if you...

- Are a patient of the East of England or North Thames Cleft Service
- Are aged between 16 and 25 years
- Have undergone corrective jaw (orthognathic) surgery between 6 months and 3 years ago

¹⁰ May 2017_version 2_IRAS ID: 211335

- OR have decided not to have corrective jaw (orthognathic) surgery between 6 months and 3 years ago

What will taking part in this research involve?

It will involve providing some basic information about yourself and taking part in a confidential interview. The interview will last between 60 to 90 minutes, will be conducted by myself and will take place at a time and location that suits you (either face-to-face at your local cleft clinic or at home or on the telephone). With your consent (agreement) the interview would be audio-recorded and then transcribed (written out).

During the interview I will invite you to talk about your role in the decision making process for corrective jaw (orthognathic) surgery, how you found this, who else was involved, and how a decision was reached. I would also like to hear about whether the cleft team did, or could do, anything more to help prepare and support you in making treatment decisions.

What will happen to the information I provide?

Your interview will be anonymised (so you are not identifiable). Yours and other people's stories will then be considered in detail to try and understand what the process of decision making for corrective jaw (orthognathic) surgery is like for young people with a cleft. This is called analysis. With your consent I would like to share my analysis from the study with other professionals and researchers by publishing the findings, so they and future patients can benefit from your expertise. If you are interested, you will be able to see a summary of the findings.

In line with the Data Protection Policy the information you provide (including your non-identifiable data) will be stored securely for 10 years in an encrypted file, on a password protected computer at the University of East Anglia.

What are the benefits and risks of taking part?

By taking part you will be helping us to understand more about your experiences and you may be able to help other people going through the decision making process have a positive experience of care.

Although I hope you will find the interview enjoyable, there is a small chance that you may be upset as it will involve discussing personal experiences. Ethics committees have to review all planned studies before they can go ahead; the purpose of this is to protect people who take part in research by making sure they do not come to any harm and to ensure that the research is of potential value to science and society. This research project has received Health Research Authority (HRA) approval and favourable opinion from the London – Brent Research Ethics Committee (REC). [REC reference: 17/LO/0817; IRAS project ID: 211335]

What else do I need to know?

Your participation is voluntary – it is up to you whether you want to take part or not. If you decide not to, the care you receive from the cleft team will not be affected. If you would like to take part, and then you change your mind, you would be able to withdraw from the research, without giving a reason, before or during the interview. You would also be able to withdraw all or part of the information you provide up to one week after the interview has taken place. After one week, analysis will have taken place

and the interview data will be anonymised and we will no longer know which answers were yours.

If you decided you no longer wanted to take part in the study and chose to withdraw, this would not affect the care you receive from the cleft team.

Unfortunately, we are unable to reimburse travel expenses so the interview will be arranged at a convenient location. However, as a thank you for giving up your time to take part you will receive a £10 voucher.

Confidentiality

All information you provide during the interview will be kept confidential. However, if you share something in your interview about harming yourself or others then this would need to be shared with your cleft team, to make sure you and others are safe; however, this would be discussed with you first.

Your GP will be notified that you have taken part in this research and it will also be recorded on your hospital patient file that you have participated in research, however no details about what you say will be shared, unless there is a risk to yourself or others (see above).

After the interview any information which identifies you will be changed. A fake name (pseudonym) of your choice will be used to protect your identity in any data or quotes used in publications.

Interested?

If you are still interested would like to be interviewed to share your experiences then we can arrange a date and time for your interview to take place. Please ask me any questions you may have about participating in this research.

How to contact us

If you have any queries, or wish to know more, please contact me via email:

m.acum@uea.ac.uk. You can also contact my supervisor Judith Young

Judith.Young@uea.ac.uk.

Thank you very much for considering taking part.

Complaints

If you wish to make a complaint about any aspect of this research please contact

Professor Ken Laidlaw (Doctorate in Clinical Psychology Programme Director), via

email K.Laidlaw@uea.ac.uk.

Appendix 11. Participant consent form

CONSENT FORM

‘Deciding about corrective jaw (orthognathic) surgery’

Please read the statements below and place your initials in the box if you agree.

I confirm that I have read and understood the participant information sheet (dated, version.....) about this research study.	
I have had the chance to ask questions and have these answered.	
I understand that my participation in this research is voluntary.	
I agree to being interviewed and my interview being audio recorded.	
I understand I can withdraw from the research, without giving a reason, and that this will not affect the care / treatment I receive.	
I understand that once my interview data has been anonymised and entered into analysis it can no longer be withdrawn. Therefore, I have <u>one week</u> after the date of my interview to request withdrawal of my interview data. After one week anonymised data will still be used in the study.	
I understand that the researcher will contact my GP and make a note on my hospital file to indicate I have taken part in this research study.	
I understand and agree for my anonymised interview data to be used in publications.	
I understand that if the researcher is concerned about mine or someone else’s safety then information may be shared with the clinical team, however this will be discussed with me first.	
I agree to take part in this research.	

Participant name:

Signed:

Date:

Researcher name:

Signed:

Date:

I have chosen a pseudonym (fake name) [] for use where I am quoted in the analysis and any publications. However, I understand this may not be able to be used, and if so, I will be given another pseudonym.	
I would like to receive a summary of the results of this research	
If so, please provide contact details below: Name: Email: Postal address:	

This research project has received Health Research Authority (HRA) approval and favourable opinion from the London – Brent Research Ethics Committee (REC). [REC reference: 17/LO/0817; IRAS project ID: 211335]

Appendix 12. Demographic information form**ABOUT YOU: DEMOGRAPHIC INFORMATION FORM****‘Deciding about corrective jaw (orthognathic) surgery’**

In order to learn about the range of people taking part in this research, we would be grateful if you could answer the following questions. All information provided is anonymous. Please either write your answer in the space provided, or circle the answer (or answers) that best apply to you.

How old are you?yearsmonths
Please indicate your gender	Female Male Other (please specify.....)
How would you describe your racial/ethnic background?	White British / White other (please specify.....) Asian / Asian British Black / Black British Mixed ethnicity (please specify.....) Other ethnic group (please specify.....) Prefer not to say
Level of education obtained	GCSE A-Level / BTEC / Other (please specify.....) Degree: Undergraduate / Masters / Doctoral
Employment status	Student: Full-time / Part-time Employed: Full-time / Part-time Unemployed
What type of cleft do you have?	Unilateral cleft lip (right / left) Unilateral cleft lip and palate (right / left) Bilateral cleft lip Bilateral cleft lip and palate Submucous cleft palate

How many surgeries have you had in relation to your cleft?	1-3 4-6 6 + (please specify how many.....)
How old were you when you made a decision about whether or not to have orthognathic surgery?	Age :
Have you ever had a session with the Clinical Psychologist within the cleft team?	Yes No Not sure
If yes, how many times have you met with the Clinical Psychologist?	Once 2-3 times 3-6 times 6 times or more (please specify.....) Not sure

Thank you for taking the time to complete this form.

Appendix 13. Example of coded transcript extract

Line no.	Transcript	Initial coding
	I: So, we're obviously going to be talking about corrective jaw surgery, so it'd be helpful to sort of know your understanding of what it is, to make sure it matches with what I've been told.	
14-18	P: So I don't know all the medical terms and words-- my jaw, bottom jaw was further forward than my upper jaw which obviously it's supposed to be the other way round. And essentially they moved the top one forward slightly and the bottom one back-- Quite a lot and yeah. That was about it. Seemed to take ages and then I had a massive swelling for about a month afterwards.	long process
	I: What do you mean when you say it took ages?	
20-26	P: I just felt like I was just waiting and waiting for it to happen like- I think the decision making process was quite a long one—when, I always just thought there's a reason that the medical professionals are recommending it so I should do it and I was-- my Mum always said I was quite laidback, probably too laid back. Like they were all worried about it and I just sort of said "yeah fine" do whatever they say this and that. Yeah that's about it really. I think I had it done December last year. So December [year].	long process being recommended - ought to have it going along with it (laidback)
	I: So you said the decision making process was quite a long one, do you remember when you first came to hear about it?	
30-37	P: I don't remember the first point of contact where it was 'you could have this'. But it's always sort of been an option as far as long I can remember really. I always knew from a young age, well not a young age, probably like 12 or 13 I knew that I was going to need, or not need, I was gonna be offered some sort of surgery. At that point they probably did say it was the jaw surgery but I was at an age where I just let Mum doing all the listening and I'll find out when, I'll cross that bridge when I come to it so, I can't really put my finger on exactly when I was told unfortunately but, I can't remember not being aware of possibly having an operation.	always on cards - further surgery less interested when younger
	I: Was the optional nature of having it, was it, did it feel like an option for you?	
60-65	P: Yeah definitely. It was always an option, to the point where I would sort of say I want you to tell me should I have or should I not but obviously they're not in a position to say that they just have to-- give the positives and negatives or the pros and cons and let you make your own decision. And it was always reiterated from everyone like my parents my extended family whoever I spoke to, it	optional nature of surgery clear wanting to have decision made for me - more direct guidance provision of info - pros and cons

	was always sort of driven into me it is an option and if I don't want to do it I don't have to.	
	I: So it felt quite optional from the kind of medical team--	
67-71	P: Yeah, at no point did I think I'm gonna have to have this. But at the same time I would've probably preferred it, because I don't know if I felt qualified enough to make a decision when-- like there'd be times where I'd come home thinking, and it's an hour and a half journey or whatever to [hospital] I'd come back thinking like please just tell me - it's better for you if you have or its better for you if you don't have it.	wanting to have decision made for me - more direct guidance
	I: Did you, before it was mentioned as an option, did you kind of think 'I really want to do something about my jaw' or--	
75-81	P: Not, very rarely, I think when you grow up with it you just assume it's normal don't you. So, it was never really anything that-- knocked my confidence hugely or made me think that I had to have it done. Every now and then you get a-- a side angle of yourself where your jaw's, your bottom jaw's hanging out [laughs] and you think that's weird but-- that was very rare so I never really thought I want this and I want this tomorrow, it needs to be done, I was in no great rush to have it done for any reason.	Accepted self - 'normal for me' didn't want surgery before mentioned - didn't see need
	I: Ok that's interesting. So it sounds like when it was talked about, I guess I'm just wondering how you felt, I mean do you remember the context that it was talked about in?	
85-96	P: Well, I had, I can't really I'm sure I had appointments with the orthodontist and stuff then I'd have separate appointments to talk about the surgery so I'd know going in that was why I was going there. And sometimes I'd be in a room with a psychologist, an orthodontist, a surgeon a nurse and speech therapist. And you'd just be sat there like this [<i>hunches shoulders together to make himself smaller</i>]	intimidation - large MDT meetings
	I: How was that to be in a room with that many people?	
	I think it must have first happened when I was 11 or 10 or and at that point I was quite intimidated and my Mum spoke to them and it didn't happen again after that for a while. And then when it happened again I spose you just expect it and it wasn't a problem. It wasn't, obviously it wasn't intentionally intimidating everyone would introduce themselves and say this is why I'm here, I'm gonna be taking notes if that's ok. But I think it's just at a young age really, more than anything.	

	I: Did it change, because I'm guessing that there was kind of those multi-professional meetings as you got older as well, how did it feel as you got older?	
100-6	P: I understood it more. I understood that I needed them there, whereas before I would think I just want to see my normal orthodontist because he's who I see every time, why do these strangers have to be there. Whereas, like I said at that age, they would have introduced themselves to me and I would have been on a different planet I wouldn't really have taken it in. So once I understood it wasn't a problem really, and I knew that like certain questions would be-- my answers would be important to different people, so I'd have to I'd just speak to him or her.	Preferred familiarity of usual professional Understanding more as got older
	So I guess, as it was being talked about more and more, did you have lots of questions about it or?	
128-30 133	No. No. I've always just had the opinion that if the medical professionals suggest it then- why not. But I do remember-- Mum would have, Mum asking quite a lot of questions. Yeah. They'd ask any questions, and I'd just look over to my Mum and...wait.	being recommended - ought to have it letting parent take responsibility - not ready?

Appendix 14. Example coding and theme development

Theme	Subthemes	Example codes
Theme 1. Awareness of difference	<i>Dissatisfaction and desire for change</i>	Feeling different Self-conscious Social pressure to fit in Bullied / teased Low self-acceptance Concern about others opinions Wanting to improve self (appearance / life) Further surgery always on cards Aware of nose Aware of jaw OS as long-term goal
	<i>It's my normal</i>	Felt ok with how was Accepted by others Mention of surgery created doubt about appearance and difference It's my normal Unawareness of jaw Drawing attention to difference
Theme 2. Committing to the process	<i>It's a long process</i>	Long process Unprepared for length of process Final milestone Burden of treatment
	<i>Making sacrifices</i>	Taking a gap year to have OS Burden on parents Other priorities Impact on life
Theme 3. Others facilitating decision making	<i>Provision of reliable information.</i>	Focus on benefits Doing own research Technical focus Wanting to know about effect on me Unprepared for aftereffects of surgery Relationship with professionals Asking questions Wanting to speak to similar others
	<i>Talking it through</i>	Time and space Talking to parents – processing information Treated as an adult Parental support Giving responsibility Value of psychology Feeling supported Wish to talk to peers
Theme 4. Responsibility on my shoulders	<i>Feeling informed, it's up to me</i>	My face, my decision Weighing up pros and cons Ability to ask questions

	Considering alternative options
	Feeling in control
	Doing own research
<i>Uncertainty about responsibility</i>	Becoming independent
	Burden of decision
	Fear of responsibility
<i>Going along with it</i>	Passive role
	Surgery being recommended
	Agreeing at various stages
	OS as standard part of treatment
	Not ready to make a decision
	Acting in my best interests

Appendix 15. Supplementary participant quotes to support derived themes

Theme Subtheme	Quote	Analytic comments
Theme 1: Awareness of difference <i>Dissatisfaction and desire for change</i>	Yeah, because you're concerned about your appearance. That's probably the main factor, and why I wanted to have this surgery done. So I was just wondering if there's anything they could do to improve my appearance. (<i>Gareth</i>)	Some YP had a prior interest and awareness of further surgery and this was a means to improving their appearance.
	I knew about the nose stuff and wanted to have that (<i>William</i>)	There was evident discontent with other aspects of facial appearance and YP wanted surgery for these.
	I was definitely always very aware of getting my nose sorted-- particularly as that was massive issue for me [...] because I was so wanting to have my nose, in my view corrected, the idea then that there was this massive bit of surgery to come in between [...] It was just, Oh there's something else we've got to do before I can get my nose done (<i>Sarah</i>)	
Theme 1: Awareness of difference <i>It's my normal</i>	It wasn't something that I registered that I needed. So it's not something that I looked into. Just cos obviously cos-- that's all I'd known in a way so I hadn't known for example-- one of the examples that was used was people not being able to take their top teeth over their bottom teeth. I wasn't able to do that, but because I never really knew about it, it wasn't something I questioned. (<i>Connor</i>)	Young people unaware of or unperturbed by their jaw alignment, as it was something they had adjusted to and had therefore become their 'normal'.
	I never thought about my jaw. I didn't like, I wasn't bothered at all. I didn't really care too much that my bite wasn't how it was mean-a be, it's just like near enough (<i>Bobby</i>)	
	I think when you grow up with it you just assume it's normal don't you. So, it was never really anything that-- knocked my confidence hugely or made me think that I had to have it done (<i>Nathan</i>)	
	This is something I've had time and time again, talking to my friends about, my nose or my jaw or any part that sort of followed the cleft lip and palate, was that they really didn't notice. [...] And I had that, that was the response every single time from everyone, whether I was just speaking to people who were trying to be particularly	As well as being unaware of the jaw personally, one YP explained how his friends also had not noticed any difference in various aspects of his appearance until

	nice to me or not, I don't know, but close friends and even my girlfriend started sort of saying the same, so I think by that point I did believe it when people said it's not something they really did notice, until it was pointed out. <i>(Mike)</i>	they were pointed out, which suggests that others were accepting.
Theme 2: Committing to the process <i>It's a long process</i>	I thought it would be-- in my view a bit of a hassle in a way. You know more appointments, more operations [...] especially as I knew, I would be going to Uni, I did think there's going to be more, kind of continuing the-- continuing the appointments and stuff rather than I suppose if I would have said no it would have been an end to the appointments and more just check-up stuff. <i>(Mike)</i>	Further treatment was initially viewed as an inconvenience in context of other life priorities. This emphasizes the burden of cleft treatment and the impact on YP's lives.
	This is kind of like the final kind of tick to tick off a successful client-- [...] It felt like the final stage but the first stage of the final stage, if that makes sense <i>(Patrick)</i>	Many YP viewed OS as the 'final milestone' that they wanted to achieve in order to reach the end of the treatment pathway.
Theme 2: Committing to the process <i>Making sacrifices</i>	I also had to balance Uni and A-Levels. If was to delay it then I'd be like-- dig into my time at Uni. Cos when I had my surgery I took a gap year, in order to recover from it. <i>(Lara)</i>	A number of YP made sacrifices in terms of taking a gap year in order to have the OS.
Theme 3: Others facilitating decision making <i>Provision of reliable information</i>	I think I needed someone to explain on like an informal level, whereas when you're in with the proper professionals [laughs] they use a lot of technical terms <i>(Patrick)</i>	The importance of language was highlighted with YP finding the use of technical language unhelpful in aiding their involvement in the decision making process.
Theme 3: Others facilitating decision making <i>Talking it through</i>	I think family and friends made a point of pointing out more of the positive side of it than anything else. But still with this whole kind of, "It's definitely your decision. It's entirely your decision but I do think it'd be really worth it". [...] So I think their kind of positive encouragement about it was the push to making decision in a way <i>(Mike)</i>	Many YP mentioned their perception that both professionals and parents showed a positive bias towards surgery, and for YP who were uncertain this influenced their decision to undergo surgery.
	The important part was having the time to think about questions to get back to them about. The doctors and team provided me	Having time and space to weigh up information, develop

	<p>enough time to ask those questions, think on it, and make a decision. <i>(Mike)</i></p>	<p>questions and reach a decision without feeling rushed was important to YP.</p>
	<p>I never felt forced into making a snap decision or, I was always told– in your own time and when you’re ready to make a decision then you make one <i>(Nathan)</i></p>	
	<p>Then she [Mum] knew it was my own decision, so when I asked her - shall I? she only just said that she was worried but you know “don’t ask me to make your choice”. [laughs] I was just a bit oh ok! Maybe I should just like step up and you know be an independent person. <i>(Lara)</i></p>	<p>In talking the decision through with parents YP found it empowering when others reinforced that the decision was the YP’s and that they would support their decision.</p>
	<p>He [Dad] didn’t like tell me either way, he was just like whatever you decide it’s up to you. But he obviously, he backed me, like I knew he was gonna back me whatever I did <i>(Bobby)</i></p>	
	<p>It was just me and my parents just sort of discussing it and then them leaving it to me to essentially decide, whatever I wanted to do they would obviously support me with so. It was just really useful as I said to just talk it through and explain like my decision process behind it rather than yeah just leaving it myself to decide what I wanna do. <i>(Thierry)</i></p>	
<p>Theme 4: Responsibility on my shoulders <i>Feeling informed – It’s up to me</i></p>	<p>Ultimately it's your life and you need to decide if you really want to get it done <i>(Gareth)</i></p>	<p>YP felt strongly about being involved and making their own decisions</p>
	<p>I was always quite keen to know kind of the whole plan and understand what was happening rather than just like get operated on. I think I probably had quite a good understanding of all of it when I came to be making the decision. <i>(William)</i></p>	<p>Feeling adequately informed appeared related to whether YP felt ready to decide.</p>
	<p>They gave me a leaflet. That was alright [laughs]. But if anything I just took to the internet to research it yeah. It was just really about what to expect, recovery process just to prepare myself you know for side effects afterwards. [Then] it didn’t seem as scary as it did not knowing about it yeah. <i>(Lara)</i></p>	<p>A number of participants mentioned doing their own research to feel better informed and feel more confident about their decision, this was particularly the case for aspects of surgery some felt underprepared for,</p>

		such as recovery and aesthetic outcomes.
<p>Theme 4: Responsibility on my shoulders <i>Going along with it</i></p>	<p>Well I was led to believe that you're not going to be unhappy [...] they'd obviously made it an option because they thought it would be a good option, which I sponse was the reassuring thing about it [laughs]. <i>(Patrick)</i></p> <hr/> <p>I think just like I said cos they were so positive about it, and they knew what the outcome would be [...] I thought like obviously they knew what was best for me. <i>(Tom)</i></p>	<p>A perception that OS was being presented as an option for a reason was common across YP's narratives and suggest trust in professionals to act in their best interests.</p>
<p>Theme 4: Responsibility on my shoulders <i>Uncertainty about responsibility</i></p>	<p>You're your own person you know suddenly you don't have to have like someone making the whole decision for you, in a sense – kind of grew up a bit more, matured. Cos before I was very dependent on my parents, you know I ask them for opinions you know and I'd take everything that they said into account. But then afterwards when I decided you know, if I was to make that choice I'd be more mature you know. <i>(Lara)</i></p>	<p>The opportunity to make a decision was a developmental milestone for some, marking the start of independence from parents.</p>