

Manuscript Number: BJOMS-D-18-00063R1

Title: A qualitative study identifying issues affecting quality of life
in adult patients with craniofacial anomalies

Article Type: Full Length Article

Keywords: Quality of Life, Craniofacial anomalies, Patient Perceptions,
Framework Analysis, Psychosocial

Corresponding Author: Miss Semina Visram, BDS; MJDF; MClintDent; MOrth:

Corresponding Author's Institution: Eastman Dental Institute; UCL

First Author: Semina Visram, BDS; MJDF; MClintDent; MOrth:

Order of Authors: Semina Visram, BDS; MJDF; MClintDent; MOrth;; Daljit
Gill, MSc; FDSRCS; FOrthRCS; Justin T Shute, MBBS; MRCPsych; MSC; Susan
J Cunningham , PhD; FDSRCS; MOrthRCS

Manuscript Region of Origin: UNITED KINGDOM

Abstract: Objective: To identify key issues affecting the quality of life
(QoL) of adult patients with craniofacial anomalies.

Method: This was a qualitative prospective study, using in-depth, semi-
structured interviews. Ten patients who fulfilled the inclusion criteria
were recruited during their attendance at the adult craniofacial clinic
at the Eastman Dental Hospital, University College London Hospitals NHS
Foundation Trust. Interviews ceased when no new themes were arising. A
thematic framework analysis was used to identify major themes and
subthemes relating to QoL in adult craniofacial patients.

Results: Of those interviewed, variation existed and, although some
positive aspects were reported, the themes which emerged were mainly
negative. Analysis identified 8 main themes, one of the major themes
being emotional issues. Within this theme, subthemes related to teasing,
bullying and abuse, as well as low mood, anxiety, depression and self-
harm.

Conclusions: Participants experienced a range of impacts as a result of
their craniofacial conditions and also expressed the need for further
emotional support. Healthcare professionals involved in the treatment of
these patients should be aware of these issues and advise where and how
further support may be accessed.

Qualitative study to identify issues affecting quality of life in adults with craniofacial anomalies

Abstract

Our objective was to identify key issues that affect the quality of life (QoL) of adult patients with craniofacial anomalies. We designed a qualitative prospective study using in-depth, semi-structured interviews. Ten patients who fulfilled the inclusion criteria were recruited during their attendance at the Adult Craniofacial Clinic at the Eastman Dental Hospital, University College London Hospitals NHS Foundation Trust. Interviews ceased when there was nothing further to discuss. A framework analysis was used to identify themes that related to QoL. Opinions varied and, although some were good, the eight main themes that emerged were mainly negative. In one of the eight, emotional issues, subthemes included teasing, bullying and abuse, as well as low mood, anxiety, depression, and self-harm. Participants experienced a range of feelings as a result of their craniofacial conditions and expressed the need for further emotional support. Healthcare professionals involved in their treatment should be aware of these issues and give advice about how to access further support.

Keywords: Quality of Life, Craniofacial anomalies, Patient Perceptions, Framework Analysis, Psychosocial

Introduction

The development of the cranium and facial bones may be affected by various conditions that have an impact on physical function, and can affect social and psychological well-being and quality of life (QoL).¹ Patients with facial disfigurement may be socially stigmatised and find it difficult to adapt to certain situations.² They may also have poor body image,³ low self-esteem, and depend on adults more than their counterparts.⁴ However, in contrast, Brantley and Clifford⁵ found a heightened sense of self-esteem among patients with clefts compared with others, and suggested that this may be the result of a patient's successful coping strategies and management of a potentially restrictive and debilitating condition.

Patients with craniofacial conditions may undergo numerous operations, often from a young age. Although good technical outcomes of surgery are reported, the psychological aspects associated with such conditions may result in the expectations and goals of surgeons and patients being different, and some people are dissatisfied with the outcome.⁶ Traditionally, outcomes of operations have been evaluated objectively - for example, using clinical photographs, and anatomical and radiographic measurements, but although these remain important, they are not sufficient on their own.⁷ It may be argued that the success of an intervention should be judged by the improvement in the patient's QoL,⁸ and it is therefore important to investigate this aspect of care.

Papers about the psychological impact of living with craniofacial conditions suggest that further research is necessary in this area, particularly among adults.^{1,2,6} The World Health Organization (WHO) has suggested the need for more evidence-based research into psychological issues and the development of QoL in patients with craniofacial conditions.⁹

The aim of this study was therefore to investigate the key issues that affect QoL in adult patients with craniofacial anomalies using a qualitative approach to gain valuable information regarding their perceptions.

Patients and methods

Study design

We designed a prospective qualitative study using in-depth interviews. The study was approved by the National Research and Ethics Service Committee, South West - Frenchay (REC reference number 13/SW/0273). Written consent was obtained from all patients, including consent to record interviews digitally with a dictaphone.

Subjects

A number of studies have investigated the QoL of patients with orofacial clefts,^{7,10,11} so we have focused on other craniofacial conditions. Participants were recruited from the Adult Craniofacial Clinic at the Eastman Dental Hospital, University College London Hospitals NHS Foundation Trust over the period October 2013 - June 2014. Patients over the age of 16 years, with a craniofacial condition (as defined by their treatment being managed in this clinic) were included. Those with orofacial clefts, or severe learning difficulties that would preclude an interview, were excluded.

Before the interviews, a framework was created to improve the generalisability of the results, and to ensure that key characteristics were reflected within the sample, including age, sex, ethnicity, and type of craniofacial condition. This was followed as closely as possible, although difficulties in recruiting patients meant that it could not be strictly adhered to.

Interviews

In-depth, semi-structured interviews were conducted at the Eastman Dental Hospital by one of the authors (SV) who used a topic guide developed specifically for the study. SV was a clinician trained in the techniques of in-depth interviewing, which involved exploring relevant issues in an open, supportive, and relaxed manner. Because of the sensitive nature of the topics being discussed, training was also provided in how to deal appropriately with difficult situations. Participants were interviewed away from the clinic, with no time constraints, and interviews were concluded when the participant had no further comments to make.

The interviews were immediately transcribed verbatim using Microsoft Word[®], and each one was coded to maintain anonymity and confidentiality.

Analysis

After the interviews had been transcribed, the themes and subthemes were identified using a framework approach to data management.¹² This matrix-based analytical method (popularised by the National Centre for Social Research (NatCen) and widely used by qualitative researchers) allows for easy comparison of interviews,¹³ and rigorous and systematic management of the data.¹²

Transcripts were read several times by two of the researchers (SV and SJC) to allow familiarisation with the raw data. Key phrases were highlighted and coded, and from this, several themes emerged.

Results

Demographics

Ten participants were recruited (Table 1). Interviews ranged in length from 15 to 45 minutes with an overall duration of 32 minutes. The data were organised using an Excel[®] workbook with nine individual spreadsheets: one recorded the demographic details and the remaining eight the main themes that emerged from the interviews. The themes and corresponding subthemes are shown in Table 2. In this paper we will focus on the “emotional issues” theme and its associated subthemes, as it raised concerns that are relevant to all clinicians who work in the area.

We have used quotes to illustrate the issues raised, including the participant’s number (for example, P1 indicates Participant 1), and added explanatory comments where appropriate. The interviews provided a large amount of data but, for the sake of brevity, we have used only a small number of quotes to illustrate each subtheme.

Emotional issues

Subtheme 1: A sense of being different

Several participants mentioned that they were aware that they were different from other people.

“I always knew I didn’t really fit in. I always knew there was something not right...not normal (laughs uncomfortably)... different. You just always wanted to be the one that just blended in, especially when you stand out, you just want to be that boring person in the background that doesn’t get noticed, and that was never me.” (P2)

Subtheme 2: People asking questions or making comments

Some interviewees described how people would ask questions or make unpleasant comments. Some, however, said that they thought that people were being inquisitive and that people were interested in them and their condition.

People say stuff. I hear the students sometimes talking and look at you in a different way.”

(P10)

Subtheme 3: Negative feelings about appearance

Several participants discussed negative feelings regarding their appearance and explained how they felt less attractive than others.

“I just don’t think I’m pretty, I don’t think I’m beautiful, I don’t think anything like that.”

(P3)

“I can’t think of anything positive. I just don’t see any positive side to it so I don’t really understand people that say they do, just the whole thing is just horrible.” (P6)

Subtheme 4: Effects of the condition on personality

The perception of their condition and the associated effects that it had on their personality varied. Although some thought that their condition enabled them to cope with difficult situations, others thought that it adversely affected their confidence, and that this was reflected in their personality and social interactions.

“It’s made me a lot stronger. I’m quite a strong person.” (P2)

“I was generally not very social. I don’t really enjoy people’s company all that much.” (P4)

Subtheme 5: Teasing/bullying

Issues regarding teasing and bullying were common. The interviewees often recalled negative remarks and encounters, and many felt that they were targeted because of their condition.

This affected them emotionally and mentally, and resulted in a lack of confidence and self-esteem.

“I was called names like ‘wonky face’... stuff like that.” (P2)

“They used to call me ‘flat face’ and ‘bobble lips’, and they would say why didn’t your family just get rid of you.....” (P3)

“...they would say things like you will always be alone because someone would have to be weird to find you attractive and things like that.” (P6)

“It [name calling] would make me feel so sad and upset and my self-esteem would just go really low.” (P10)

“I’ve had really horrible things said to me... I’ve had people say it’s the mark of the devil.” (P6)

Subtheme 6: Abuse from others (physical/ verbal/ psychological)

Because of their severity, some experiences were categorised as abuse rather than teasing or bullying. Participants discussed difficult times when they were subjected to physical, verbal, and psychological abuse, and the emotional effects these had on them.

“I didn’t have any friends, I was bullied, every word under the sun, I was called everything. I was attacked and all sorts ...I used to be kicked, punched, slapped, scratched ... by all the other kids. I had food and drinks poured all over me, like crisps poured down my back and then punched into the back of me.” (P7)

Subtheme 7: Issues relating to anxiety, depression, self-harm, or suicide

The consequences of these situations ranged from patients saying that they “felt down”, to anxiety, depression, self-harm, and suicidal thoughts. Roughly half the participants said that they had been depressed, had self-harmed, or had suicidal thoughts, at some point in their lives.

One participant admitted to self-harming and had been admitted to hospital for depression. Others had been prescribed medication for anxiety or depression, or both, and another discussed suicidal thoughts.

“...I was self-harming and I was admitted to hospital for 2 weeks with depression through self-harming and things. I was on all different medications.” (P7)

“Well there’s too many issues with me really... there’s too many problems... some days I feel like, oh gosh... I just don’t want to be in this world anymore.” (P3)

Subtheme 8: Understanding/acceptance

In contrast to the situations described above, some participants accepted their condition and did not feel such levels of negativity towards it. Understanding and accepting themselves and their condition were important in making them feel comfortable about themselves.

“You don’t want people to feel sorry for you; you want them to understand what you’re going through.” (P9)

“It’s all-right, because it’s life, because your life is not the same as others, so obviously you have different problems and different things so that’s why I don’t feel no problem, I feel OK.” (P8)

Subtheme 9: Positive aspects of the condition

The final theme also addressed some positive aspects. Nearly half of those interviewed said that the condition had helped to strengthen their personality and character, and they appreciated that it had made them a more interesting person.

“It’s made me a lot stronger. I’m quite a strong person... people say that and it’s made me take things as they come.” (P2)

Discussion

The method used in this study was successful, as most participants discussed aspects of their QoL in detail and also covered a range of sensitive issues. Interviews varied in length, partly because of the nature of the person being interviewed, but overall they lasted for about 32 minutes, which allowed for a great deal of information to be discussed.

We recruited patients until no new themes became apparent, although it is accepted that this concept of “theoretical saturation” is unpredictable. It is important to recognise that it is not necessary to have a large sample in this type of qualitative research, as data on incidence or prevalence, and statistical tests, are not needed.¹² However, a sample of 10 patients is small to reach “theoretical saturation”, and taking into account the complex nature of the issues discussed, further areas of concern may have been raised if more patients had been interviewed.

Although participants discussed both positive and negative aspects of their conditions, most comments were negative. Several people explained that their adverse feelings were a result of their physical appearance and how others perceived them, and many felt that this had a detrimental effect on their everyday life and their personality.

These findings are similar to those in other studies that identified the negative impact of social stigmatisation encountered by these patients. In a questionnaire-based study, Geirdal et al¹⁴ found that increased psychological distress and poorer QoL were associated with certain craniofacial conditions. Pruzinsky¹⁵ reported that social and psychological challenges adversely affected the QoL of patients with severe craniofacial deformities and their families, and Sarwer et al⁶ found lower levels of self-esteem and QoL in adults with craniofacial anomalies compared with a control group. The overall psychological impact on our patients is similar to that found by Singh and Moss¹⁶ in their questionnaire-based study of 112 craniofacial patients (including those with clefts), in which they found greater appearance-related distress in comparison with a control group.

Most participants had experienced some form of teasing and bullying during their lives, and many felt that their condition made them more susceptible. They recognised and discussed the distressing impact this had on themselves and their families. Carroll and Shute¹⁷ found that patients with craniofacial conditions often had to face aggressive behaviour and bullying at school, but also noted that those without such conditions were victimised and suffered emotional distress. As part of their study, Edwards et al¹⁸ conducted in-depth, semi-structured interviews with adolescents who had craniofacial conditions, and identified seven main themes of which one comprised stigma and isolation.

Participants also discussed low mood, anxiety, depression, self-harm, and suicide, and several were currently seeking help or had sought it in the past. Previous studies have acknowledged that an increased number of patients with facial deformities have psychiatric diagnoses and behavioural problems, and although patients with orofacial clefts were not included in the

current study, similarities are evident in this group.¹⁰ Christensen et al¹⁹ found that patients with congenital defects had an increased risk of death, not only during childhood but also in adulthood, and found that suicide rates were higher in both sexes affected than in the general Danish population.

This raises important concerns about the psychological evaluation of patients with craniofacial conditions.²⁰ Our study shows that mental health issues are important. All members of the craniofacial team should be aware of them and direct patients who are at risk to the support that they require. Teams that treat adults would benefit from the expertise of a dedicated psychologist or mental health professional as in clinics that treat children and adolescents. If this is not feasible, such issues should be raised with patients who should then be directed to services such as IAPT (Improving Access to Psychological Therapies) in the UK.

Several charities and organisations also help to support patients and families. While they undoubtedly have an important role, it is important to find out whether patients can access them appropriately or feel comfortable doing so. Patients can be told about confidential support and advice from healthcare specialists or charity organisations such as Headlines and Changing Faces in the UK.^{21, 22} Information can be distributed, for example, by medical or dental practitioners, specialists on multidisciplinary teams, or teachers. Posters, leaflets, and information on television and radio can also raise awareness of these organisations.

While most of the discussions focused on negative aspects of their conditions, a small number of patients spoke about positive aspects. In their questionnaire-based study, Marcusson et al¹¹ found that patients with cleft lip and palate had also adjusted well to life

despite their condition. Raposo-Amaral et al²³ also compared patient-reported QoL between patients with Apert and Crouzon syndromes, and found that it was satisfactory in both groups.

Our study has limitations. The participants had a variety of conditions and future work could focus on specific disorders to improve the homogeneity of the results. Also, because of the nature of the problem, some patients had undergone physical treatment from a young age whereas some had had treatment later in life, and others were still awaiting some aspects of care. However, as the rarity of many of these disorders makes recruitment difficult, multicentre studies should be considered in future. Nevertheless, despite individual variation, the participants described similar experiences regardless of their condition.

The study may have benefitted if we had interviewed some older patients. The age range of 16-25 years covers a time when there can be important changes in life, and this might limit the conclusions that can be drawn.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patients' permission

The study was approved by the National Research and Ethics Service Committee South West - Frenchay (REC reference number 13/SW/0273). Patients were given information leaflets and they all gave written informed consent.

Note to author

I have omitted reference 21, as it was the same as reference 11, and renumbered accordingly. I have also altered the order of the last 3 references to put them into numerical order.

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Table 1.

Participants' details.

	No. (n = 10)
Sex:	
Men	4
Women	6
Mean (range) age (years)	20 (16-25)
Ethnicity:	
White ethnic group	7
Black African-Caribbean	1
Asian or Asian British	2
Craniofacial condition:	
Crouzon syndrome	3
Apert syndrome	2
Other complex craniosynostosis	1
Parry-Romberg syndrome	1
Hemifacial microsomia	1
Binder syndrome	1
Treacher-Collins syndrome	1

Table 2.

The main themes and subthemes that emerged after analysis of the data.

Main themes	Subthemes
Physical issues	Facial and non-facial issues; dressing differently/disguising features
Emotional issues	A sense of being different; people asking questions/ making comments; negative feelings about appearance; effects of the condition on personality; teasing/ bullying; physical/ verbal/ psychological abuse; anxiety, depression, self-harm or suicide; understanding/ acceptance; positive aspects of the condition
Education	Type of education; interactions; achievements; sense of academic unfulfilment/ underachievement; current career/ employment
Effects of treatment	Type of treatment and reasons for; positive and negative physical and emotional effects; time in hospital
Coping strategies	Crying/ getting upset; support network/ talking to people; acceptance; other; psychiatric/ pharmacological support; expressing a need for increased support
Relationships	Parents/step parents/siblings/partners (positive and negative relationships); friends; other family members affected by the condition; relationship with other people who have the condition; online communication
Information seeking	Methods of seeking information; awareness of information; expressing need for increased availability
Future concerns and issues	Future jobs/ employment; living situations; relationships; children

Orthodontic Department
Birmingham Community Healthcare NHS Trust
Birmingham Dental Hospital & School of Dentistry
5 Mill Pool Way
Edgbaston
Birmingham
United Kingdom
B5 7EG

Dear Mr Mitchell

Ms. Ref. No.: BJOMS-D-18-00063

A qualitative study identifying issues affecting the Quality of Life in adult craniofacial patients

The British Journal of Oral & Maxillofacial Surgery

Very many thanks for reviewing the above article and thank you to the referees for their positive and constructive feedback. The reviewers' comments have been addressed as detailed at the end of this letter. Please also find attached a copy of our resubmission with all amendments highlighted in red.

If there are any changes which you would like us to consider, please do not hesitate to let us know.

With many thanks

Yours sincerely

Semina Visram

Reviewer #1:

This is a very interesting study and concisely written.

In the title rather than 'adult craniofacial patients' as this is a very broad term it might be better to use (as in the abstract) 'adult patients with craniofacial anomalies'.

This has been addressed on page 1, and highlighted in red.

It is qualitative paper using framework method of analysis. Item saturation needs to be confirmed at just 10 patients as this is on the small number side but might be representative if the patients have similar 'quality of life' issues. However the authors state that 'great variation existed amongst the individuals regarding issues affecting QoL'.

This has been addressed on page 1 and page 4. Participants were interviewed until no new themes were arising. Although there was variation between patients, the major themes were consistent and the wording has therefore been amended to "variation existed" rather than "great variation existed".

Eight main themes were identifies. Although not necessarily surprising, the major theme of emotion is perhaps unexpected to be so prominent given the adult population, support given on childhood / adolescence, adaption and coping over time. Also there are national associations and charitable organisations, so one assumes they are wide failing to support the child and family if there is still so much distress in adulthood.

This comment has been addressed on page 14. It is not the intention of the authors to be critical to those organisations which offer valuable support and we hope the editor is happy with the wording utilised.

The authors state that 'a number of studies currently exist investigating the quality of life in individuals with the most common craniofacial condition, oro-facial clefting'. Comparison with the findings of these papers and the hypothesis as to why other craniofacial conditions might be different, would be helpful.

This comment has been addressed on page 13. We hope this adequately answers the question.

The selection process of the 10 patients is not clear not how the final number was reached. Also an outline of previous surgeries and planned surgeries. Were they all at the end of treatment. Did age and number of treatments make a difference.

Patients were recruited over a specified time period according to the inclusion/exclusion criteria specified. It has been acknowledged that recruitment was difficult due to the rarity of the conditions; this is mentioned in the method and discussion on pages 4 and 14 respectively. Due to the small numbers, subgroup analyses were not undertaken and we selected not to give too much information regarding treatment in case patients felt their anonymity had not been completely protected by giving details of the condition, age, gender and type of treatment.

Some of the interviews seemed very short e.g 15 minutes. With such apparent intensity of concerns it is surprising that these could be adequately articulated and illustrated in such a short time frame.

This comment has been addressed on page 11. The mean interview time was 32 minutes and this allowed a great deal of information to be discussed. We hope this adequately answers the question.

Are there any other references that might help put the findings of this study into context?

Further discourse including some of the references suggested has been provided on pages 12-14. We hope this adequately answers the question.

Reviewer #2:

The authors are to be congratulated on tackling a difficult topic in craniofacial surgery: their identified emotional themes - and the patient evidence to substantiate them - clearly highlight that this is an area of craniofacial surgery that must be addressed. In this regard there is much that can be learned from how these issues have been tackled in cleft surgery but it would be interesting to note any differences. The results of the paper make for interesting reading but the introduction and discussion would benefit from a more thorough, and up to date, discourse of the available literature of which there is plenty. As they currently stand, the introduction and discussion do not do the results - and the methods to obtain them - justice, rendering the paper an insipid read when it could be so much more.

Broadly speaking I find the following areas disappointing:

1. This is THE era of PROMS. With so much having been published in the area of Facial Disfigurement (Karen Wing-Riff, Angela Pusic having authored landmark papers) it is astonishing to me that the reference list is so out of date without mention of these important papers which cite research on the emotional issues upon which many validated PROMS questionnaires have been based. This is particularly so with regard to the research on cleft-related emotional issues. I would be interested to know whether these issues differ in any way from those identified in pre-existing literature investigating the craniofacial population. Further discourse has been provided on pages 12-14. The systematic review undertaken by Klassen et al. (2012) has been included. Other more current references have also been highlighted (references pages 17-18). We have found work by Angela Pusic but this is mainly on breast reconstruction and other aspects of facial aesthetics so we have chosen not to include this. If the editor would like it to be included, we are happy to do so.

2. "Literature regarding the psychological impact of living with craniofacial conditions suggests that further research is necessary in this area, particularly regarding the adult population" - this should ideally be referenced

This comment has been addressed on page 3 with references 1, 2 and 6. We hope this adequately answers the question.

3. "Patients were excluded if they presented with severe learning difficulties or reduced cognitive development (due to the nature of the interviews undertaken) and those with orofacial clefting"

Was formal IQ testing undertaken? How was reduced cognitive development ascertained? To what extent is this considered a confounder in the assessment of "key issues affecting the quality of life" - is there any evidence for this?

This comment has been addressed on page 4, the patients who were excluded were primarily those with learning difficulties which were apparent to the clinical team. We hope this adequately addresses the concern.

4. "Participants were recruited until no new themes appeared to be arising, at a point when 'theoretical saturation' is sometimes said to have been achieved"

Regarding an issue as complex as quality of life, it is difficult to believe that after just 10 participants, theoretical saturation was already achieved. Was this really the reason? I very much appreciate the difficulty in recruiting patients and the huge time investment involved in this study - were these contributory factors that should be honestly declared? I note that the small sample size is defended later in the article - twice.

We appreciate the reviewer's comments regarding this and this has been addressed on pages 11 and 14.

5. A question: content analysis is often achieved with software? Was this the case and is so could this be declared in the methodology please?

The data was organised using a classic thematic framework approach (utilizing an Excel[®] workbook with 9 spreadsheets). No software was used as there was no requirement in a study of this size.

6. Discussion

This - much like the introduction - is wholly disappointing. For such impactful results (given such a small sample size) I would suggest that an evidence-based discourse on the prevalence of emotional issues amongst these patients be foremost in the discussion, followed by the need for further enquiry and surgeon / team awareness. Instead the discussion begins with a paragraph that ought to be placed in the methodology section.

We thank the reviewer for their comments and we have added some further discussion. We hope this adequately addresses the comments made.

7. "Pruzinsky discussed the social and psychological challenges faced by patients " PruzAnsky?

The reference has been rechecked and the spelling 'Thomas Pruzinsky' is correct.

8. In addition, adult craniofacial teams would benefit from the inclusion of a psychiatrist or mental health professional so that they can offer their advice and expertise in the same way that most child and adolescent teams are able to do so.

Do you mean psychologist or psychiatrist? Cleft multidisciplinary teams should include a dedicated cleft-specific psychologist - not psychiatrist.

This comment has been addressed on page 13. The research team believes that the important aspect is to have a mental health professional on the team and this could be a psychologist or liaison psychiatrist (as on some orthognathic and plastic surgery MDTs).

BRITISH JOURNAL OF ORAL & MAXILLOFACIAL SURGERY

Author contribution

Manuscript Title ___ A qualitative study identifying issues affecting the Quality of Life in adult craniofacial patients ___

Please provide details in the table below of each author(s) contribution to the submitted manuscript

AUTHORS	Conception and design of study/review/case series	Acquisition of data: laboratory or clinical/literature search	Analysis and interpretation of data collected	Drafting of article and/or critical revision	Final approval and guarantor of manuscript
Semina M. Visram , MOrthRCS; MClintDent; BDS	Involved in study design	Involved in literature searches	Responsible for interviewing subjects, data collection and analysis	Responsible for drafting and revising the article	Approved final version and guarantor of manuscript
Daljit Gill , MSc; FDSRCS; FOrthRCS	Involved in the initial study concept and design	Involved in supervising the literature searches	Responsible for approving topic guide, interviewing training and data analysis	Responsible for revising and reviewing the article	Approved final version of manuscript
Justin T Shute , MBBS; MRCPsych MSc ³	Involved in study design	Involved in overseeing the study and providing psychiatric support	Responsible for approving topic guide, interviewing training and data analysis	Responsible for revising and reviewing the article	Approved final version of manuscript
Susan J. Cunningham , PhD; FDSRCS; MOrthRCS ¹	Involved in the initial study concept	Involved in supervising the literature searches	Responsible for approving topic guide, interviewing training and data analysis	Responsible for revising and reviewing the article	Approved final version of manuscript

Dear Mr Mitchell

Ms. Ref. No.: BJOMS-D-18-00063

A qualitative study identifying issues affecting the Quality of Life in adult craniofacial patients

The British Journal of Oral & Maxillofacial Surgery

Very many thanks for reviewing the above article and thank you to the referees for their positive and constructive feedback. The reviewers' comments have been addressed as detailed at the end of this letter. Please also find attached a copy of our resubmission with all amendments highlighted in red.

If there are any changes which you would like us to consider, please do not hesitate to let us know.

With many thanks

Yours sincerely

Reviewer #1:

This is a very interesting study and concisely written.

In the title rather than 'adult craniofacial patients' as this is a very broad term it might be better to use (as in the abstract) 'adult patients with craniofacial anomalies'.

This has been addressed on page 1, and highlighted in red.

It is qualitative paper using framework method of analysis. Item saturation needs to be confirmed at just 10 patients as this is on the small number side but might be representative if the patients have similar 'quality of life' issues. However the authors state that 'great variation existed amongst the individuals regarding issues affecting QoL'.

This has been addressed on page 1 and page 4. Participants were interviewed until no new themes were arising. Although there was variation between patients, the major themes were consistent and the wording has therefore been amended to "variation existed" rather than "great variation existed".

Eight main themes were identified. Although not necessarily surprising, the major theme of emotion is perhaps unexpected to be so prominent given the adult population, support given

on childhood / adolescence, adaption and coping over time. Also there are national associations and charitable organisations, so one assumes they are wide failing to support the child and family if there is still so much distress in adulthood.

This comment has been addressed on page 14. It is not the intention of the authors to be critical to those organisations which offer valuable support and we hope the editor is happy with the wording utilised.

The authors state that 'a number of studies currently exist investigating the quality of life in individuals with the most common craniofacial condition, oro-facial clefting'. Comparison with the findings of these papers and the hypothesis as to why other craniofacial conditions might be different, would be helpful.

This comment has been addressed on page 13. We hope this adequately answers the question.

The selection process of the 10 patients is not clear not how the final number was reached. Also an outline of previous surgeries and planned surgeries. Were they all at the end of treatment. Did age and number of treatments make a difference.

Patients were recruited over a specified time period according to the inclusion/exclusion criteria specified. It has been acknowledged that recruitment was difficult due to the rarity of the conditions; this is mentioned in the method and discussion on pages 4 and 14 respectively. Due to the small numbers, subgroup analyses were not undertaken and we selected not to give too much information regarding treatment in case patients felt their anonymity had not been completely protected by giving details of the condition, age, gender and type of treatment.

Some of the interviews seemed very short e.g 15 minutes. With such apparent intensity of concerns it is surprising that these could be adequately articulated and illustrated in such a short time frame.

This comment has been addressed on page 11. The mean interview time was 32 minutes and this allowed a great deal of information to be discussed. We hope this adequately answers the question.

Are there any other references that might help put the findings of this study into context?

Further discourse including some of the references suggested has been provided on pages 12-14. We hope this adequately answers the question.

Reviewer #2:

The authors are to be congratulated on tackling a difficult topic in craniofacial surgery: their identified emotional themes - and the patient evidence to substantiate them - clearly highlight that this is an area of craniofacial surgery that must be addressed. In this regard there is much that can be learned from how these issues have been tackled in cleft surgery but it would interesting to note any differences. The results of the paper make for interesting reading but the introduction and discussion would benefit from a more thorough, and up to date, discourse of the available literature of which there is plenty. As they currently stand, the introduction and discussion do not do the results - and the methods to obtain them - justice, rendering the paper an insipid read when it could be so much more.

Broadly speaking I find the following areas disappointing:

1. This is THE era of PROMS. With so much having been published in the area of Facial Disfigurement (Karen Wing-Riff, Angela Pusic having authored landmark papers) it is astonishing to me that the reference list is so out of date without mention of these important papers which cite research on the emotional issues upon which many validated PROMS questionnaires have been based. This is particularly so with regard to the research on cleft-related emotional issues. I would be interested to know whether these issues differ in any way from those identified in pre-existing literature investigating the craniofacial population Further discourse has been provided on pages 12-14. The systematic review undertaken by Klassen et al. (2012) has been included. Other more current references have also been highlighted (references pages 17-18). We have found work by Angela Pusic but this is mainly on breast reconstruction and other aspects of facial aesthetics so we have chosen not to include this. If the editor would like it to be included, we are happy to do so.

2. "Literature regarding the psychological impact of living with craniofacial conditions suggests that further research is necessary in this area, particularly regarding the adult population" - this should ideally be referenced

This comment has been addressed on page 3 with references 1, 2 and 6. We hope this adequately answers the question.

3. "Patients were excluded if they presented with severe learning difficulties or reduced cognitive development (due to the nature of the interviews undertaken) and those with orofacial clefting"

Was formal IQ testing undertaken? How was reduced cognitive development ascertained? To what extent is this considered a confounder in the assessment of "key issues affecting the quality of life" - is there any evidence for this?

This comment has been addressed on page 4, the patients who were excluded were primarily those with learning difficulties which were apparent to the clinical team. We hope this adequately addresses the concern.

4. "Participants were recruited until no new themes appeared to be arising, at a point when 'theoretical saturation' is sometimes said to have been achieved"

Regarding an issue as complex as quality of life, it is difficult to believe that after just 10 participants, theoretical saturation was already achieved. Was this really the reason? I very much appreciate the difficulty in recruiting patients and the huge time investment involved in this study - were these contributory factors that should be honestly declared? I note that the small sample size is defended later in the article - twice.

We appreciate the reviewer's comments regarding this and this has been addressed on pages 11 and 14.

5. A question: content analysis is often achieved with software? Was this the case and is so could this be declared in the methodology please?

The data was organised using a classic thematic framework approach (utilizing an Excel[®] workbook with 9 spreadsheets). No software was used as there was no requirement in a study of this size.

6. Discussion

This - much like the introduction - is wholly disappointing. For such impactful results (given such a small sample size) I would suggest that an evidence-based discourse on the prevalence of emotional issues amongst these patients be foremost in the discussion, followed by the need for further enquiry and surgeon / team awareness. Instead the discussion begins with a paragraph that ought to be placed in the methodology section.

We thank the reviewer for their comments and we have added some further discussion. We hope this adequately addresses the comments made.

7. *"Pruzinsky discussed the social and psychological challenges faced by patients "*
PruzAnsky?

The reference has been rechecked and the spelling 'Thomas Pruzinsky' is correct.

8. *In addition, adult craniofacial teams would benefit from the inclusion of a psychiatrist or mental health professional so that they can offer their advice and expertise in the same way that most child and adolescent teams are able to do so.*

Do you mean psychologist or psychiatrist? Cleft multidisciplinary teams should include a dedicated cleft-specific psychologist - not psychiatrist.

This comment has been addressed on page 13. The research team believes that the important aspect is to have a mental health professional on the team and this could be a psychologist or liaison psychiatrist (as on some orthognathic and plastic surgery MDTs).

Title: A qualitative study identifying issues affecting the quality of life in adult patients with craniofacial anomalies

List of Authors:

Semina M. Visram, MOrthRCS; MCLinDent; BDS ¹

(Specialty Registrar in Orthodontics)

Daljit Gill, MSc; FDSRCS; FOrthRCS²

(Consultant in Orthodontics)

Justin T Shute, MBBS; MRCPsych MSc³

(Liaison Psychiatrist)

Susan J. Cunningham, PhD; FDSRCS; MOrthRCS¹

(Professor/ Honorary Consultant in Orthodontics)

¹ University College London Eastman Dental Institute, UK

²Great Ormond Street NHS Foundation Trust and Eastman Dental Hospital, University College London Hospitals Foundation Trust, London, UK

³Eastman Dental Hospital, University College London Hospitals Foundation Trust, London, UK

Corresponding Author Contact Information:

Miss Semina Visram

Orthodontic Department

Birmingham Community Healthcare NHS Trust

Birmingham Dental Hospital & School of Dentistry

5 Mill Pool Way

Edgbaston

Birmingham

B5 7EG

Tel: 07885415459 (UK)

semina.visram@nhs.net

This manuscript has been presented at the following meetings:

1. Craniofacial Society of Great Britain and Ireland, Annual Scientific Conference; London, UK; April 15, 2015.
2. British Orthodontic Society, University Teachers Group session at the International Orthodontic Congress, London, UK; September 27, 2015.

Running head: Quality of life in craniofacial patients

Financial disclosure and products page

None of the authors declare any commercial or financial associations for the submitted work; or financial relationships with any organisation that might have an interest in the submitted work or other relationships or activities that could appear to have influenced the submitted work.

A qualitative study identifying issues affecting **quality of life in adult patients with craniofacial anomalies**

Abstract

Objective: To identify key issues affecting the quality of life (QoL) of adult patients with craniofacial anomalies.

Method: This was a qualitative prospective study, using in-depth, semi-structured interviews.

Ten patients who fulfilled the inclusion criteria were recruited during their attendance at the adult craniofacial clinic at the [REDACTED] Dental Hospital, [REDACTED]

[REDACTED]. **Interviews ceased when no new themes were arising.** A **thematic** framework analysis was used to identify major themes and subthemes relating to QoL in adult craniofacial patients.

Results: **Of those interviewed, variation existed and,** although some positive aspects were reported, the themes which emerged were mainly negative. Analysis identified 8 main themes, one of the major themes being emotional issues. Within this theme, subthemes related to teasing, bullying and abuse, as well as low mood, anxiety, depression and self-harm.

Conclusions: Participants **experienced a range of impacts as a result of their craniofacial conditions and also** expressed the need for further emotional support. Healthcare professionals involved in the treatment of these patients should be aware of these issues and advise where and how further support may be accessed.

Keywords: Quality of Life, Craniofacial anomalies, Patient Perceptions, Framework Analysis, Psychosocial

Introduction

The development of the cranium and facial bones may be affected by various craniofacial conditions. In addition to having an impact on a patient's physical functioning, these conditions may also contribute to social and psychological well-being and affect quality of life (QoL).¹ It has been reported that patients with facial disfigurement may suffer from social stigmatisation and find it difficult to adapt to certain situations.² They may also demonstrate poor body image,³ lower self-esteem and increased dependency on adults compared with their counterparts.⁴ However, in contrast, Brantley and Clifford⁵ found a heightened sense of self-esteem amongst cleft patients, compared with non-cleft individuals and suggested that this may be due to a patient's successful coping strategies and management of a potentially restrictive and debilitating condition.

Patients with craniofacial conditions may undergo numerous surgical procedures, often from a very young age. Although, high technical outcomes of surgery are reported, some individuals remain dissatisfied with the results of their treatment.⁶ The psychological aspects associated with such conditions may result in a difference in expectations and goals between the surgeon and patient. Outcomes following surgical interventions have traditionally been evaluated using objective measurements, for example using clinical photographs, anatomical measurements and radiographic measurements. Although these traditional methods of determining outcomes remain important, they are not sufficient on their own.⁷ It may be argued that the success of an intervention should be determined by an improvement in the quality of life of that individual,⁸ and it is therefore important that this aspect of care is also investigated.

Literature regarding the psychological impact of living with craniofacial conditions suggests that further research is necessary in this area, particularly regarding the adult population.^{1,2,6} In addition, the World Health Organisation (WHO) suggests greater need for evidence-based research into psychological issues and development of quality of life measures for patients with craniofacial conditions.⁹

The aim of this study was therefore to investigate the key issues affecting quality of life in adult patients with **craniofacial anomalies** using a qualitative approach in order to provide a method of social inquiry and gain valuable information regarding patients' perceptions.

Subjects and Methods

Study Design

This was a prospective qualitative study utilising in-depth interviews. The study was granted favourable ethical opinion by the National Research and Ethics Service Committee South-West Frenchay (REC reference number 13/SW/0273). Written consent was obtained from all patients, including consent to digitally record interviews with a Dictaphone.

Subjects

As a number of studies currently exist investigating the quality of life in individuals with the most common craniofacial condition, oro-facial clefting,^{7,10,11} this study focused on other craniofacial conditions. Potential participants were recruited from the Adult Craniofacial Clinic at the [REDACTED] over the period from **October 2013 to June 2014**. The inclusion criteria were those patients over the age of 16 years, with a craniofacial condition (as defined by their treatment being managed in the Adult

Craniofacial Clinic). Patients were excluded if they had oro-facial clefting, or had severe learning difficulties which would preclude an interview being undertaken.

Prior to undertaking in-depth interviews, a purposive sampling framework was created in order to enhance the generalisability of the results. This aimed to ensure that key characteristics within the population were reflected within the sample, including age, gender, ethnicity and type of craniofacial condition. This was followed as closely as possible, although difficulty in recruiting patients meant that this could not be strictly adhered to.

Interviews

In-depth, semi-structured interviews were conducted at the [REDACTED] by one researcher (SV), using a topic guide developed specifically for this study. SV was a clinician trained in the techniques of in-depth interviewing, which involved exploring issues of relevance in an open, non-leading and relaxed manner. Due to the sensitive nature of the topics being discussed, training was also provided in dealing with difficult situations in the most appropriate manner. Participants were interviewed away from the clinical environment, with no time constraints and interviews were terminated when the participant had no further comments to make. Participants were recruited until no new themes were arising, a point at which 'theoretical saturation' is sometimes said to have been achieved.

The interviews were immediately transcribed verbatim, using Microsoft Word[®] and each interview was coded in order to maintain anonymity and confidentiality.

Analysis of the Interviews

Following transcription of the interviews, a **thematic** content analysis was used to identify themes and subthemes, with a framework approach to data management.¹² This is a matrix based analytical method popularised by the National Centre for Social Research (NatCen) and allows easy comparison of the interviews.¹³ It is a method widely used by qualitative researchers and allows rigorous management of the data in a systematic fashion.¹²

Transcripts were read several times, by two of the researchers (SV and SJC), to allow familiarisation with the raw data. Key phrases were highlighted and coded and this led to the emergence of a number of themes.

Results

Demographics

Ten participants were recruited, based on the purposive sampling framework, and their demographics can be seen in Table 1. Interview times ranged in length between 15 minutes and 45 minutes, with an average interview time of 32 minutes. The data was organised using an Excel[®] workbook with 9 **individual** spreadsheets, one recorded the demographic details and the remaining 8 represented the main themes which emerged from the interviews. The themes and corresponding subthemes are shown in Table 2. This article will focus on the ‘emotional issues’ theme and its associated subthemes. This theme raised concerns which were felt to be relevant to all clinicians working in this area.

Quotes are used to illustrate the issues raised, participant numbers are included (e.g. P1 indicates Participant 1) and explanatory comments have been added where appropriate. **The**

interviews provided a large amount of data but for the sake of brevity in this paper, it is only possible to use a small number of quotes to illustrate each subtheme.

Main Theme: Emotional Issues

Subtheme 1: A sense of being different

Several participants mentioned that they had an awareness of being different in comparison with other people.

“I always knew I didn’t really fit in. I always knew there was something not right ...not normal (laughs uncomfortably)... different. You just always wanted to be the one that just blended in, especially when you stand out, you just want to be that boring person in the background that doesn’t get noticed and that was never me.” (P2)

Subtheme 2: People asking questions or making comments

A number of the interviewees described how people would ask questions or make comments that were of a negative nature. Some, however, described how they felt people were being inquisitive and said that they felt it showed that people were interested in them and their condition.

“People say stuff. I hear the students sometimes talking and look at you in a different way.” (P10)

Subtheme 3: Negative feelings about appearance

A number of the participants discussed negative feelings regarding their appearance and described how they felt less attractive than others.

“I just don’t think I’m pretty, I don’t think I’m beautiful, I don’t think anything like that.”

(P3)

“I can’t think of anything positive. I just don’t see any positive side to it so I don’t really understand people that say they do, just the whole thing is just horrible.” **(P6)**

Subtheme 4: Effects of the condition on personality

The perception of their condition, and the associated effects on personality, varied amongst the participants. Although some felt their condition empowered them so that they were able to cope with difficult situations; others felt it had a negative effect on their confidence and this was reflected in their personality and their social interactions.

“It’s made me a lot stronger. I’m quite a strong person.” **(P2)**

“I was generally not very social. I don’t really enjoy people’s company all that much.” **(P4)**

Subtheme 5: Teasing/ Bullying

Issues regarding teasing and bullying were common amongst those interviewed. The interviewees often recalled negative remarks and encounters, and many felt they were targeted because of their craniofacial condition. This resulted in participants feeling as though they lacked confidence, being aware of reduced self-esteem and reporting that they were emotionally and mentally affected.

“I was called names like ‘wonky face’... stuff like that.” (P2)

“They used to call me ‘flat face’ and ‘bobble lips’, and they would say why didn’t your family just get rid of you.....” (P3)

“...they would say things like you will always be alone because someone would have to be wierd to find you attractive and things like that.” (P6)

“It [name calling] would make me feel so sad and upset and my self-esteem would just go really low” (P10)

“I’ve had really horrible things said to me... I’ve had people say it’s the mark of the devil” (P6)

Subtheme 6: Abuse from others (Physical/ Verbal/ Psychological)

In some instances, the severity of the experiences described, were categorised as abuse rather than teasing or bullying. Participants discussed difficult times when they were subjected to physical, verbal and psychological abuse and the negative emotional effects this had on them.

“I didn’t have any friends, I was bullied, every word under the sun I was called everything, I was attacked and all sorts ...I used to be kicked, punched, slapped, scratched ... by all the other kids. I had food and drinks poured all over me, like crisps poured down my back and then punched into the back of me.” (P7)

Subtheme 7: Issues relating to anxiety, depression, self-harm or suicide

The negative consequences of these situations ranged from the patients saying they “felt down”, to anxiety, depression, self-harm and suicidal thoughts. Approximately half of the participants said that they had been affected by depression, had self-harmed or had suicidal thoughts at some point in their life.

One participant admitted to self-harming and had been hospitalised for depression, others had received medication for anxiety and/or depression and another participant discussed their suicidal thoughts.

“...I was self-harming and I was admitted to hospital for 2 weeks with depression through self-harming and things. I was on all different medications.” (P7)

“Well there’s too many issues with me really...there’s too many problems... some days I feel like oh gosh... I just don’t want to be in this world anymore.” (P3)

Subtheme 8: Understanding/ Acceptance

In contrast to the situations described above, some participants discussed how they were able to accept their condition and did not feel such levels of negativity towards it. Understanding and acceptance of themselves, and their condition, was important in making them feel comfortable about themselves.

“You don’t want people to feel sorry for you; you want them to understand what you’re going through.” (P9)

“It’s all-right, because it’s life, because your life is not the same as others, so obviously you have different problems and different things so that’s why I don’t feel no problem, I feel ok.” (P8)

Subtheme 9: Positive aspects of the condition

The final theme also addressed some more positive aspects. Of those interviewed, nearly half said that the condition had helped to strengthen their personality and character and they appreciated how it had made them a more interesting person.

“It’s made me a lot stronger. I’m quite a strong person... people say that and it’s made me take things as they come.” (P2)

Discussion

The methodology used **in this study** proved successful, as most of the participants discussed aspects of their QoL in detail, and also covered a range of sensitive issues. **There was a variation in interview times which was partly related to the nature of the individual being interviewed, however, an average interview time of 32 minutes allowed a great deal of information to be discussed.**

Patients were recruited until it was felt that no new themes were arising; although it is accepted that this concept of ‘theoretical saturation’ is unpredictable. It is important to recognise that it is not necessary to have a large sample size in qualitative research of this type, as it is not necessary to gather data with regards to incidence/prevalence or to test statistical significance of statements.¹² **However, a sample size of 10 patients is small to reach ‘theoretical saturation’ and further areas of concern may have been raised if additional interviews had been undertaken, taking into account the complex nature of the issues discussed.**

Analysis of the data identified 8 main themes to emerge (Table 2) and the emotional issues affecting these patients have been illustrated further in this paper. The results show that although participants discussed both positive and negative aspects of their conditions, the majority of comments were of a negative nature. A number of individuals discussed how they felt that the adverse feelings they had were as a result of their physical appearance and how

others perceived them, many felt that this consequently led to a detrimental effect on their everyday lives and their personality.

These findings were similar to other studies which identified the negative impact of social stigmatisation encountered by patients who have craniofacial deformities. In their questionnaire-based study, Geirdal *et al.*¹⁴ showed that patients had increased psychological distress and reduced quality of life associated with certain craniofacial conditions.

Pruzinsky¹⁵ discussed the social and psychological challenges faced by patients with severe craniofacial deformities and concluded that these patients, and their families, experienced a negative impact on their quality of life as a result. Similarly, Sarwer *et al.*⁶ reported lower levels of self-esteem and quality of life in adults with craniofacial anomalies compared with a control group. The overall psychological impact on patients reported in this study is supported by Singh and Moss¹⁶ in their questionnaire-based study of 112 craniofacial patients (including patients with cleft), where they found greater appearance related distress in comparison with a control population.

The majority of participants had experienced some form of teasing and bullying during their lives, and many felt that they were more susceptible to this due to their craniofacial condition. The participants recognised and discussed the distressing impact this had on themselves and their families. Similar results have been discussed in other research; Carroll and Shute¹⁷ found that those patients with craniofacial conditions were more frequently targeted with aggressive behaviours and school-based peer-victimisation. However, they also noted that those without a craniofacial condition also suffered frequent victimisation and emotional distress. As part of their study, Edwards *et al.*¹⁸ conducted in-depth semi-structured

interviews with adolescents who had craniofacial conditions and identified stigma and isolation as one of their 7 main themes.

Participants also discussed low mood, anxiety, depression, self-harm and suicide and several were currently seeking help or had sought help in the past. Previous studies have acknowledged the increased number of patients with facial deformities, who have psychiatric diagnoses and behavioural problems. **Although patients with orofacial clefts were not included in the current study, similarities are evident with this cohort of patients¹⁰.**

Christensen *et al.*¹⁹ found that patients with congenital defects had an increased risk of mortality, not only during childhood but also in adulthood and found that suicide rates were higher in both males and females than in the general Danish population.

This raises important concerns surrounding the psychological evaluation of craniofacial patients.²⁰ It is apparent from this study that mental health issues are significant, and all members of the craniofacial team should be aware of these concerns and direct patients who are at risk through the appropriate channels so that they can gain the support they require. In addition, adult craniofacial teams would benefit from the expertise of a dedicated **psychologist or mental health professional** in the same way that most child and adolescent clinics do. If this is not feasible, clinics should raise the issues with patients and signpost them to services where further support is available such as the IAPT (Improving Access to Psychological Therapies) system in the UK.

A number of charities and organisations currently exist to help support patients and families. Whilst these organisations undoubtedly play an important role, it is important to assess if patients are always able to access them appropriately or feel comfortable doing so. Patients

can be made aware of confidential support and advice from healthcare specialists or charity organisations, such as Headlines and Changing Faces in the UK^{23, 24}. This information may be distributed to patients in a number of settings, for instance via their medical or dental practitioner, by specialists on multi-disciplinary teams, or by teachers at schools. Provision of information through posters, leaflets, television and radio may also increase awareness of such organisations.

Whilst the majority of the discussions focused on negative aspects of craniofacial conditions, a small number of patients were able to draw on positive aspects of their condition. Similarly, in their questionnaire-based study, Marcusson *et al.*²¹ found that patients with cleft lip and palate had formed good life adjustments in spite of their condition. Raposo-Amaral *et al.*²² also compared patient reported quality of life between Aperts and Crouzon patients and found that both had a satisfactory quality of life.

There are limitations associated with the study. The participants had a variety of conditions and future work could focus on specific craniofacial conditions in order to enhance the homogeneity of the results. In addition, due to the nature of the conditions some patients had undergone physical treatment from a very young age whereas others had treatment later in life and others were still awaiting some aspects of care. However, the rarity of many of these conditions makes recruitment for such research difficult and suggests that multi-centre studies should be considered in the future. Nevertheless, despite individual variation, similar experiences were described by the participants, regardless of their condition.

The study may also have benefitted from interviewing and gathering data from some older participants, the age range of 16-25 years is a time when important life changes may occur and this might limit the conclusions which can be drawn.

Conclusions

From the semi-structured interviews, 8 main themes were identified in relation to the quality of life of adults with craniofacial conditions. Although a number of common themes were identified, variation existed with regards to past experiences and the effects of the craniofacial condition on an individual's quality of life. A number of important issues were raised by participants regarding emotional difficulties they had experienced and many indicated that further support would be beneficial. Healthcare professionals involved in the treatment of individuals with craniofacial conditions should be familiar with the issues affecting these patients and be able to direct them to additional support which is available.

Conflict of Interest

None of the authors declare any commercial or financial associations for the submitted work.

Ethics statement/confirmation of patient permission

The study was granted favourable ethical opinion by the National Research and Ethics Service Committee [REDACTED] ([REDACTED]). Subjects involved gave informed consent. Patient information leaflets were provided and written consent was obtained for all participants that were involved in the study.

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Table Legends

Table 1: Demographics of Interview Participants

Table 2: The main themes and subthemes following analysis of the data

Table 1

Gender	Number	Age Range (Mean)	Ethnicity	Number	Craniofacial condition	Number
Males	4	16-25 (20.2)	White ethnic group	7	Crouzons syndrome	3
Females	6		Black Afro-Caribbean	1	Aperts syndrome	2
			Asian or Asian British	2	Other complex craniosynostosis	1
					Parry-Rhomborg syndrome	1
					Hemifacial microsomia	1
					Binder's syndrome	1
					Treacher-Collins syndrome	1

Table 1: Demographics of Interview Participants

Table 2

Main Themes	Physical Issues	Emotional Issues	Education	Effects of Treatment	Coping Strategies	Relationships	Information Seeking	Future Concerns and Issues												
Subthemes	Facial Issues	A sense of being different	Type of Education	Type of Treatment	Crying/ Getting upset	Parents (Positive)	Methods of seeking information	Future Jobs/ Employment												
	Non-Facial Issues	People asking questions/ making comments	Interactions	Reasons for having Treatment	Support Network/ Talking to People	Parents (Negative)	Awareness of information	Living Situations												
	Dressing Differently/Disguising Features	Negative feelings about appearance	Achievements	Physical Effects (Positive)	Acceptance	Step-Parents (Positive)	Expressing need for increased availability	Relationships												
		Effects of the condition on personality	A sense of Academic Unfulfilment/ Underachieving	Physical Effects (Negative)	Other	Step-Parents (Negative)		Children												
		Teasing/ Bullying	Current Career/ Employment	Emotional Effects (Positive)	Psychiatric/ Pharmacological Support	Siblings (Positive)														
		Abuse from others (Physical/ Verbal/ Psychological)		Emotional Effects (Negative)	Expressing a need for Increased Support	Siblings (Negative)														
		Issues relating to anxiety, depression, self-harm or suicide		Time in Hospital		Friends														
		Understanding/ Acceptance		Other Family Members affected by the condition																
		Positive aspects of the condition		Relationship with other people who have a condition																
				Partners (Positive)																
		Partners (Negative)																		
		Online Communication																		

Table 2: The main themes and subthemes following analysis of the data



The British Journal of Oral & Maxillofacial Surgery
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REFERENCE NO: ..BJOMS-D-18-0006321.....

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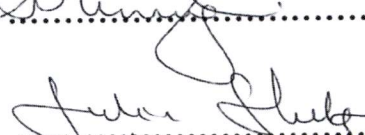
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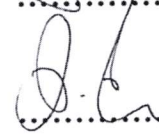
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