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

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Abstract

Our objective was to identify key issues that affect the quality of life (QoL) of adult patients with craniofacial anomalies. 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 arose. A framework method of analysis was used to identify themes that related to QoL. Opinions varied and, although some were positive, the eight main themes that emerged were mainly negative. One of the main themes was that of emotional issues. Within this theme, 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 support. Healthcare professionals involved in the treatment of these patients, 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

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Introduction

The development of the cranium and facial bones may be affected by various craniofacial conditions that have an impact on physical function, and may also contribute to social and psychological well-being and affect 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 increased dependancy on adults compared with their counterparts.[4] However, in contrast, Brantley and Clifford[5] 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, some individuals remain dissatisfied with the results of their treatment. The psychological aspects associated with such conditions may result in a difference in expectations and goals between the surgeons and patient.[6] Traditionally, outcomes of operations have been evaluated objectively – for example, using clinical photographs, and anatomical and radiographic measurements. Although these remain important, they are not

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sufficient on their own.[7] It may be argued that the success of an intervention should be judged by an 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 measures 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.

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

Patients and methods

Study design

This was a prospective qualitative study using 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 record interviews digitally with a dictaphone.

Subjects

A number of studies have investigated the QoL of patients with orofacial clefts,[7,10,11] this study has therefore 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. 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). Those with orofacial clefts, or severe learning difficulties that would preclude an interview, were excluded.

Before the interviews were begun, a purposive sampling 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 completed at the Eastman Dental Hospital by one of the authors (SV), using 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, non-leading 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 clinical environment, with no time constraints, and interviews were concluded when the participant had no further comments to make. Participants were recruited until no new themes arose, 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 to maintain anonymity and confidentiality.

Analysis

After transcription of the interviews, the themes and subthemes were identified using a framework approach to data management.[12] This is a matrix-based analytical method, that was popularised by the National Centre for Social Research (NatCen), and allows for easy comparison of interviews.[13] It is a method used by qualitative researchers and allows 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 general interview time of 32 minutes. The data were organised using an Excel© workbook with nine individual spreadsheets: one recorded

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; reasons for having treatment; 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

the demographic details and the remaining eight represented the main themes that emerged from the interviews. The themes and corresponding subthemes are shown in Table 2. This paper will focus on the “emotional issues” theme and its associated subthemes, as it raised concerns that were thought to be relevant to all clinicians working in this area.

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

A number of 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)

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 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.

Patients were recruited 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 it is not necessary to gather data with regards to incidence or prevalence, or to test the significance of statements statistically.[12] However, a sample of 10 patients is small to reach "theoretical saturation", and taking into account the complex nature of the issues discussed, it is accepted that 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 participants explained that their adverse feelings were a result of their physical appearance and how others perceived them, 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 other patients with craniofacial deformities. In a questionnaire-based study, Geirdal et al[14] found that increased psychological distress and poorer QoL were associated with certain craniofacial conditions. Pruzinsky[15] reported that social and psychological challenges adversely affected the QoL of patients with severe craniofacial deformities and their families and Sarwer et al[6] 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 Moss[16] 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 had made them more susceptible to this. They recognised and discussed the distressing impact this had on themselves and their families. Carroll and Shute[17] 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 al[18] 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 al[19] 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, similarly to the way that clinics that treat children and adolescents do. 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 al[11] found that patients with cleft lip and palate had also adjusted well to life despite their condition. Raposo-Amaral et al[23] 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 from interviewing and gathering data from 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.

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.

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.

Conflict of interest

We have no conflicts of interest.

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