

Medical stress reactions and personal growth in parents of children with a rare craniofacial condition

Abstract

Background: The birth of a child with a congenital craniofacial anomaly (CFA) can have a profound psychological impact on the family. Despite the complexity and variability of treatment for these rare conditions, few studies have been conducted into parents' emotional responses to healthcare experiences. The aim of the present study was to investigate parents' subjective experiences of their child's condition and treatment using an in-depth qualitative approach.

Methods: Individual semi-structured interviews were conducted in person or over the telephone with 48 parents of children with a range of rare CFAs. Interviews were transcribed verbatim, translated into English, and analysed using inductive thematic analysis.

Results: Participants reported physical and psychological symptoms that could be indicative of medical traumatic stress in relation to their child's diagnosis and treatment. Participants described feelings of powerlessness, and the weight of being responsible for their child's care. Yet, participants also reported that as a result of their experiences, their perspective on life had changed, and they had grown in self-confidence.

Conclusions: The findings provide insight into the complex physical and psychological effects experienced by parents in response to their child's diagnosis and medical treatment, as well as an understanding of how these experiences may also result in personal growth over time. Implementation of trauma-informed evidence-based resources should be considered in craniofacial care and future research, particularly in regard to prevention and treatment of psychological distress.

Key words: Qualitative, craniofacial, caregiver stress, medical traumatic stress, personal growth.

Introduction

A congenital craniofacial anomaly (CFA) is a comprehensive term used to describe a wide range of diagnoses, such as craniosynostosis and Treacher Collins syndrome, which may be isolated to the head and neck, or may form part of a broader genetic condition (Holmbeck & Aspinall, 2015). Prevalence rates and characteristics vary widely across the many craniofacial conditions and may affect the form and function of the head and face, as well as cognitive functioning (Buchanan et al., 2014). Further, many domains of psychological adjustment may be impacted (Feragen & Stock, 2017). Depending on the condition and associated malformations, complex multidisciplinary treatment is likely to be required throughout childhood and often into adulthood. Typically affected areas are the jaws, the mid-face, cheekbones, eyes and ears, the respiratory tract, and in severe cases a restricted growth of the skull. As a result, many patients may experience problems with regular breathing, which may also impact the quality of sleep, development of speech, chewing and eating, and symptoms from elevated intracranial pressure, such as headaches and progressive vision loss. Surgical treatment aimed at minimising the original malformation will often be recommended. Parents of children with CFAs will therefore have to cope with their child undergoing multiple treatments, and in some cases life-threatening situations, such as acute respiratory difficulties. The ever-present nature of the child's treatment course, coupled with the uncertainty of treatment protocols, may understandably be emotionally demanding for parents, and could potentially trigger stress reactions (Lim et al., 2019; Pinquart, 2018; Pope et al., 2005).

Research on children with health needs indicates that treatment demands may adversely affect the parents and increase their psychological vulnerability (Jackson et al., 2016; Lim et al., 2019), which may affect the long-term wellbeing of the family as a whole (Lim et al., 2019; Mirhosseini et al., 2015). Most parents manage to cope with stress reactions to their child's diagnosis and/or treatment, while others experience more persistent symptoms that could develop into trauma-related stress over time (Pinquart, 2018). In order to describe common reactions after challenging medical events across multiple paediatric conditions (Kazak et al., 2006), the term 'Paediatric Medical Traumatic Stress' (PMTS) has been proposed, and defined as "a set of psychological and physiological responses of children and their families to pain, injury, serious illness, medical procedures, and invasive and frightening treatment experiences" (Price et al., 2015, p.86). Traumatic stress reactions, while conceptualised as understandable, are potentially distressing physical and emotional responses

to very challenging experiences (Kassam-Adams et al., 2013), such as a medical condition that involves a threat to the child's health, or an intrusive, painful, or alarming treatment experience (Kazak et al., 2006). Traumatic stress responses may include arousal, re-experiencing, and avoidance symptoms that in some cases may become disruptive to functioning. Another closely related term is 'Post-Traumatic Stress Disorders' (PTSD). Although PMTS and PTSD are related and share some symptoms, PMTS may be present without meeting criteria for a full diagnosis of PTSD (Price et al., 2015).

The presence of stress reactions among parents of children with a medical condition has been broadly investigated. One third of parents of children who are admitted to paediatric intensive care units or who undergo surgery for a congenital disease develop acute stress reactions (Nagata et al., 2008; Balluffi et al., 2004), or intense feelings of fear and anxiety (Sjöström-Strand & Terp, 2017). Another review described frequent reports of depression, anxiety, somatic symptoms, fear of recurrence, extensive worry, and fatigue (Ljungman et al., 2014). Woolf-King and colleagues (2017) reported in their systematic review that between 30-80% of parents of children undergoing cardiac surgeries reported significant symptoms of psychological distress. Other studies have explicitly investigated the presence of PMTS or PTSD, reporting that approximately a third of parents of children with cancer or cardiac surgeries develop symptoms of PTSD (Ljungman et al., 2014; Woolf-King et al., 2017).

Qualitative research on CFAs has shed light on several psychologically demanding aspects of treatment. For example, parents have reported concerns for their child's medical and physical safety (Klein et al., 2006; Klein et al., 2010), and both parents and patients have highlighted the emotional burden of ongoing treatment (Klein et al., 2010; Roberts & Shute, 2010; Stavropoulos et al., 2011). Quantitative findings in CFAs are less clear, whereby some studies identify elevated levels of parental stress (Pope et al., 2005) while others find no differences between clinical samples and controls groups (Pinquart, 2018; Lim et al., 2019). As mentioned by Lim et al. (2019), the majority of available CFA studies are based on single suture craniosynostoses and more research is needed on more complex CFAs. To the authors' knowledge, no studies have explicitly investigated PMTS in CFAs.

The effects of traumatic stress symptoms may not be entirely negative. Some people who are exposed to traumatic events do not develop PMTS or PTSD, and some may even gain psychological benefits (Joseph &

Linley, 2006; Ljungman et al., 2014; Meyerson et al., 2011). This phenomenon is known as ‘Posttraumatic Growth’ (Seligman & Csikszentmihalyi, 2000), and describes positive psychological changes that may result from life-altering experiences. Several models of post-traumatic growth have emphasised the importance of self-reflection and the pursuit, construction, and alteration of meaning for growth to occur (Joseph & Linley, 2006). Post-traumatic growth has been theoretically described as including five domains: greater appreciation of life, improved personal relationships, greater personal strength, recognitions of new possibilities in one’s life course, and spiritual or religious growth (Tedeschi & Calhoun, 1996). Although investigation of strengths, as well as difficulties has increased in the craniofacial field in recent years (Feragen & Stock, 2017), further exploration of personal growth in the context of rare CFA diagnosis and treatment is needed, in order to provide a more balanced view of psychological adjustment. Investigations of parental growth could allow for clinical intervention and prevention work to encompass positive coping strategies and promote resilience.

Clinical knowledge about treatment demands and the limited research on caregiver stress in CFAs suggests that the investigation of stress symptoms, whether these are time-limited and moderate, or more severe and indicative of PMTS or PTSD, could be indicated in parents of children with CFAs. There is little knowledge about how difficult emotions and negative parental treatment-related experiences translate to day-to-day life in rare and complex CFAs, particularly when compared to other paediatric populations (Bally et al., 2018). Parents’ subjective interpretations of medical events continue to be one of the most powerful predictors of stress reactions across conditions (Price et al., 2015). Qualitative research can therefore provide crucial insight into the relationship between a child’s health condition, diagnostic and/or treatment related experiences, and psychological distress among parents, particularly when quantitative findings are inconclusive. Yet, the psychological impact of complex and demanding treatment on parents of children with CFAs has not been investigated in detail. This is particularly surprising given the increased complexity and variability of treatment for many of these rare conditions (Feragen & Stock, 2017). The aim of the present study was therefore to investigate parents’ positive and negative subjective experiences of their child’s CFA treatment, using an in-depth qualitative approach.

Methods

Design and Procedure

The Norwegian National Unit for Craniofacial Surgery is responsible for the centralised, multidisciplinary treatment of all patients affected by rare CFAs in the country. The unit consists of representatives with expertise from different medical specialties, such as plastic and neurosurgeons, geneticists, otolaryngologists, ophthalmologist, radiologists, speech and language therapists, and orthodontists. The multidisciplinary team does not include psychological expertise, but referrals can be made to relevant services if a need is identified. All parents attending a multidisciplinary (MDT) meeting with the craniofacial team during the study period (September 2016-October 2017) were approached to participate in this study. Prior to the MDT meeting, information about the study was sent to parents by post, containing further details about what participation in the study would entail, and key ethical information such as confidentiality and their right to withdraw. Some participants returned the consent form by post and an appointment for the interview was made over the telephone. Those who did not contact the researchers prior to their MDT meeting were approached in person at the clinic. Among the 81 parents who received information about the study, only five (6.5%) chose not to participate. Fourteen parents (17%) responded positively but were not reached when contacted for an interview appointment. Another 14 did not respond (17%), which could indicate a lack of available time, a lack of perceived relevance, or the absence of up-to-date contact information. A total of 48 parents participated, leading to a final participation rate of 59%.

Individual, semi-structured interviews were conducted with parents of children born with a range of relatively rare CFAs. Interview appointments with the 48 participants were made for the same day as the MDT meeting, or shortly after. Interviews were conducted face-to-face ($n = 14$) or over the telephone ($n = 34$) by the third author, who is a qualified clinical psychologist trained in qualitative methods, and not a member of the centralised treatment team. Interviews were 60 minutes in length on average (range = 30 to 75 minutes).

The current paper was extracted from data collected as part of a larger qualitative study. For the full study, a semi-structured interview guide aimed at investigating treatment experiences was created by the first and third authors, by drawing upon clinical experience and current literature from the craniofacial and broader health fields. Participants were asked open-ended questions and were prompted to provide more details where appropriate. An overview of the topics and questions regarding treatment experiences covered in the full

interview guide is provided in Table 1. During analysis of the larger dataset, a broad range of parental stress reactions was identified. These data were subsequently extracted from the full dataset and are presented in the current paper.

Participants

Participants in this study included 48 parents (40 mothers, 8 fathers) of 46 children (32 males; 16 females) with relatively rare CFAs. Children's ages ranged from one year to 18 years¹ (mean age = 9.5 years; 30 males). Children's conditions included Treacher Collins syndrome, Crouzon syndrome, craniosynostosis, Goldenhar syndrome, Muenke syndrome, and Apert syndrome, in addition to particularly rare genetic conditions that will not be mentioned to protect anonymity. Most participants' children were still under treatment from the multidisciplinary team, while a few were able to reflect on the treatment pathway as a whole. Some children also had an additional diagnosis of autism spectrum disorder, cognitive difficulties, and/or epilepsy. In three cases, one of the parents had the same condition as the child. All parents except four were Norwegian. A total of 70% of participants were married or living in partnership with the child's mother/father at the time of interview, while 12 parents (26 %) were divorced or had left their partner. Marital status was missing for two of the participants (4 %). Most families had two (38 %) or three children (27 %), including the child with a CFA.

Ethical Considerations

The Data Protection Office at Oslo University Hospital granted ethical approval for the current study (2016/14088). Relevant referrals or follow-up could also be arranged by the interviewer if necessary.

Analysis

This study utilised an in-depth, inductive qualitative approach. Interviews were audio-recorded, transcribed verbatim, and translated into English. Inductive thematic analysis was used to analyse the data. This approach is deemed advantageous when the aim of a study is to summarise a large body of qualitative data, particularly when existing theory and/or research literature on a given phenomenon is limited (Braun & Clarke, 2006). Analyses were carried out by the first two authors following the guidance provided by Braun and Clarke

¹ One of the children was older than 18 years. This participant had a severe developmental disorder and consequently remained in the full custody of his/her parents. Therefore, these parents were included in the sample.

(2006): (1) becoming familiar with the data; (2) identifying interesting features of the data; (3) searching for themes; (4) reviewing themes; (5) defining and naming themes; and (6) producing the report. Analysis was seen as a recursive process, and detailed notes were written throughout. Themes were subsequently chosen for their prevalence and/or their importance in relation to the research questions. Themes were discussed between all authors until full agreement was reached.

Results

Two core themes were identified. The first was ‘Psychological Distress and Traumatic Experiences,’ which included three sub-themes: ‘Powerlessness and Uncertainty,’ ‘Responsibility,’ and ‘Physical and Emotional Impact’. The second core theme was ‘Personal Growth,’ which included two sub-themes: ‘Change of Perspective’ and ‘Change in Self-Confidence’. Themes and sub-themes are outlined below using exemplar quotes and are summarised in Figure 1. All participants and their children have been given pseudonyms. In some instances, quotes have been shortened by the authors, in order to enhance clarity and focus. This is indicated by the use of (...).

Theme 1: Psychological Distress and Traumatic Experiences

The vast majority of participants reported psychologically distressing and/or potentially traumatic experiences in relation to their child's medical condition. Feelings of powerlessness and responsibility were reported, alongside several physical and psychological consequences.

Powerlessness and Uncertainty

Participants reported struggling with the uncertainty associated with their child's condition. This often began prior to the confirmation of the diagnosis:

“The ultrasound revealed problems. We were told the child would not live... and that started a heavy process” (Mary, daughter aged 4).

“It took six months before we had a diagnosis. That was the worst period of time for us, not knowing what it was (...). It didn't feel safe not to know” (Olivia, son aged 6).

“You didn't know what lay ahead (...). We didn't know what to expect” (Sarah, son aged 10).

Participants also described uncertainty in relation to their child's ongoing treatment, and their fears for their child's future:

"Nobody can give me an answer, so you have to live with the uncertainty then" (Katie, son aged 12).

"[My son] has experienced more pain and more surgery than most people will in their whole lives (...). And there is more to come" (Joe, son aged 6).

Participants reported the powerlessness they felt when they could not support their child as they would have otherwise done instinctively:

"You are put in a situation where you are not the one who knows what is best for your child (...). Other people take control, and in a way decide how your child is going to have it (...). This is difficult, because your protective instinct is so strong" (Mary, daughter, aged 4).

"It hurts so much to be a parent and not have any influence on this. I am used to fixing what is difficult, but I can't fix this" (Helena, son aged 12).

Several participants had watched their child endure one or more life-threatening events, during which they had experienced an acute lack of control:

"I went to the coffee shop, taking all the necessary medicines with me, and then my son became more and more congested. So I found the suction machine in order to clear the tracheostomy, and the battery (...) had just died. I felt more and more stressed. So I asked the man in the coffee shop for a straw, and there I was, clearing the tracheostomy with the straw and spitting on the floor. [My son] was nearly blue. [These experiences] have been really traumatic for me, as his mother" (Katie, son aged 12).

"They had to fly my son by helicopter over to the regional hospital. There wasn't any space for me with the emergency services, so I had to travel in a normal passenger plane, a few hours after birth" (Thea, son aged 3).

For other parents, the threat to their child's life had spanned a longer period of time, and the feeling of powerlessness had been ongoing:

“We were so afraid to lose him. And we couldn’t do anything (...). We were so scared all the time. So, as a result, you die a little inside, because you don’t know how to handle this” (Charlotte, son aged 5).

“I almost wrote an obituary...because I went from day to day and knew nothing. But you hoped - maybe maybe, maybe” (Susanna, son aged 15).

Responsibility

In an attempt to regain some control, participants had taken on the responsibility of researching their child's condition themselves:

“I was searching for information on the Internet about everything (...) [even] different operation techniques that could be used” (Patricia, son aged 6).

Often, this also stemmed from participants' observations that non-specialist HPs, such as midwives and paramedics lacked knowledge about their child's condition and how to treat it.

“[The midwives] just thought [my son] had broken his nose during the birth (...). But I understood immediately - ‘no, no, no, he needs help’” (Jane, son aged 10).

“I called the emergency services, but when they came they were completely dumbstruck, they had never seen a child like this” (Katie, son aged 12).

Similarly, participants described the feeling of not having their concerns about their child taken seriously by hospital staff:

“All the time it took to (...) be believed by the doctors, all this time you wear yourself out, because you don’t get the help you need and no one understands how big your problems are” (Rosie, son aged 15).

These experiences resulted in participants lacking trust in some HPs, and a constant feeling of needing to be on guard:

“I didn’t trust the nurses would be able to care for my son. So I followed them all the time, I couldn’t leave him (...). I found it so scary that someone else would care for him” (Thea, son aged 3).

“I have learned that I can’t put it all in the hands of the hospital, because they do make mistakes, they forget, they give the wrong medicines (...), so you have to constantly follow up” (Eve, son > 18 years).

Participants described how they had gradually taken on responsibility for their child's care:

“Over time, I have become an administrative director (...). It feels like I am teaching them, like I am the expert” (Katie, son aged 12).

“We can’t wait for others to do their job. They have many people waiting for their help, we only have Christopher. So we take the big job of sorting everything out ourselves” (Anna, son aged 3).

Participants expressed how this sense of responsibility weighed heavily on them as parents:

“It is difficult to be in this role, almost as a nurse or a doctor (...) and to sometimes feel like more of a health professional than you would like to” (Joe, son aged 6).

Physical and Emotional Impact

Participants recounted the physical impact that the experiences of their child’s treatment had had on them. For some, this had included acute physiological sensations:

“I was reading with the television on in the background, and suddenly I felt a rush through my whole body, and I thought, ‘wow, what happened?’ And I saw that my child’s surgeon had been featured in a [TV] programme (...). My whole body had reacted to his voice without my awareness of it.” (Lauren, daughter aged 7).

Many participants described exhaustion and insomnia, often leading to physical burnout:

“We were physically exhausted (...). We had no energy left, we couldn’t take care of ourselves (...). We didn’t sleep at all” (Anna, son aged 3).

“We were too tired to be able to care for him as he really needed (...). People admired us, but...we just stood up in the morning and did what we had to” (Eve, son > 18 years).

“I hit a wall. I couldn’t handle it anymore. I was on sick leave for years (...). You reach a point where you feel totally burned out” (Sarah, son aged 10).

Participants also described the psychological toll of their child's condition. This included feeling guilty, scared, anxious, emotionally drained, and depressed. For many, this had been apparent from the point of diagnosis:

"It hurts so very, very much to think back to the time when I considered terminating the pregnancy" (Mary, daughter, aged 4).

"It felt like an awful bubble. You are supposed to be happy because you have had a child, and then it feels totally wrong. You wonder how this is going to be, and you are scared, and you don't feel a mother's happiness... it is quite overwhelming actually (...). We just lived in a kind of survival mode" (Katie, son aged 12).

"We felt like we had tumbled down onto a different planet" (Charlotte, son aged 5).

Participants outlined the impact their child's condition had on their relationships with their partner, family members, and friends:

"Our love story did not exist anymore (...). We wanted to be together, but we just didn't know how to manage the situation" (Elsa, son aged 12).

"The first year was dreadful. [My husband and I] couldn't talk to each other because we didn't know what to say. We were just very, very scared. All the time." (Charlotte, son aged 5).

"This was a burden on the whole family. For the other kids, for me and my husband. We did not have any extra energy (...) to be what we were supposed to be for each other" (Rosie, son aged 15).

"Almost all our friends backed out, they didn't know how to handle him. They were afraid of him dying in their arms" (Elsa, son aged 12).

Additionally, participants reported that the need to sanction treatment had affected the bond between them and their child:

"It is clear that it hasn't been OK when you feel you break your child's trust in you" (Joe, son aged 6).

As a result of these physically and psychologically demanding experiences, participants believed that families should be offered professional support:

“Parents should be offered help from a psychologist (...). There are so many different phases and it is so demanding on daily life (...). I feel it is sad that this support is lacking” (Katie, son aged 12).

“The local hospital did not offer us any possibilities to talk about our feelings (...). I wish we had been offered some support as parents” (Patricia, son aged 6).

Theme 2: Personal Growth

Despite the many physical and psychological consequences of their child’s condition and treatment, the majority of participants were able to identify positive outcomes of their experiences. This included a change of perspective, and a change in self-confidence.

Change of Perspective

Participants reflected on the possibility that their situation could have been worse:

“We have seen children on the ward (...) with cancer and other life-threatening conditions. I believe we have been lucky after all” (Lauren, daughter aged 7).

“I was sure he would be completely disabled. So I feel good to see him walk and he is so wonderful (...). So I believe we will make it (...). It is difficult, but it could have been so much worse (...). We are all alive. I try to think like this” (Rosie, son aged 15).

Participants reported having a new outlook on life:

“We can’t have all the good things in life, others must have some as well (...). We have learned a lesson that life includes ups and downs” (Anna, son aged 3).

“You have to live in the moment and be aware of the small things in life” (Peter, son aged 10).

“Everything can be fixed. There is a solution for all problems (...). One phase passes, and it was only a phase” (Elisabeth, son aged 15).

Participants also noted an increased capacity for empathy for others:

“I really understand other people who struggle, more easily now than I did before” (Ivan, son aged 10).

“We all meet people who have fought a battle, and we don’t know which battle they have fought, and I believe it is important to...try to make the world a better place for them” (Mathilda, son aged 8).

Similarly, participants spoke of the process they had been through to become more accepting of their own situation and that of others:

“Over time (...) I have learned not to judge people by how they look” (Carrienne, son aged 15).

“It has been a process (...). To accept Marianne as she is, and that she will need her parents more than other children. Transforming the disappointment and sorrow into something else (...), and accepting that the world is as it is” (Mary, daughter aged 4).

Change in Self-Confidence

Participants believed their experiences had made them stronger, less afraid, and more able to cope with the challenges related to their child’s condition:

“It feels good to know that I am (...) the type of person who can take a punch” (Mary, daughter, aged 4).

“You learn a lot about yourself (...). The experience of being so low and then to manage to rise again (...). I believe I am a strong person, that I have managed this (...), that I survived” (Rosie, son aged 15).

“I can cope more easily (...), I don’t feel completely down when something happens (...). Nothing is worse than when something is wrong with your children, so when it happens (...) you know you can take most other things. You know then that you have a strength you can find when you need it” (Alice, son aged 18).

Participants also attributed their increase in confidence to their developed understanding of complex medical information:

“I have acquired a lot of medical knowledge. Staying in a hospital for a year with a sick child makes you very good at this. I almost feel like a licensed nurse without being one” (Susanna, son aged 15).

“I learned a lot (...), and I have more respect for myself now” (Anna, son aged 3).

As well as experiencing a positive change in themselves, participants believed they had become more competent parents:

“I have good knowledge of my child. I believe I am good at being his mum” (Jane, son aged 10).

“You feel kind of like a ‘superdaddy’, because of everything you have been through” (Steven, son aged 15).

In parallel, participants described the importance of being a role model for their family:

“I want my children to see that their mum copes (...). They can think: ‘my Mum, she handled it’. I hope they see that” (Mathilda, son aged 8).

Discussion

The findings of this qualitative study provide important insight into complex physical and psychological reactions that parents may experience in response to their child’s CFA and medical treatment and their potentially significant impact on everyday life and relationships, as well as an understanding of how these experiences may also result in positive changes to parents’ life perspective and perceived self-confidence over time. It is proposed that stress reactions, whether mild or severe, acute or long-lasting, could be conceptualised as normal reactions to an abnormal situation, rather than as a pathological reaction to situational cues (Frankl, 1984; Kassam-Adams et al., 2013). Still, caregiver stress may be associated with negative health outcomes, and impact the entire family system (Lim et al., 2019; Pinquart, 2018). The present study thus hopes to draw attention to parents who may struggle to cope with stress reactions, so that medical teams can identify those at risk and refer them to adequate support. The present study indicates that in order to prevent long-lasting negative impacts and promote positive outcomes, the assessment of parental stress reactions may need to be provided more systematically.

The Impact of Emotional Distress and/or Potentially Traumatic Experiences

Recent reviews have investigated the nature and prevalence of acute emotional distress and/or traumatic stress reactions in parents of children in paediatric care (Bally et al., 2018; Price et al. 2015; Ljungman et al., 2014). In the present study, participants reported physical and psychological responses to their child’s surgical interventions, medical complications, and postoperative care. These responses, which included guilt, grief, anxiety, fear, depression, physical exhaustion, or a loss of control were powerfully described, even if they had occurred as many as 15 years ago, supporting the theory of enhanced memory availability for emotional laden

events (Rubin et al., 2014). Participants' emotional and physiological reactions were most commonly triggered by their child's need for constant care (Ljungman et al., 2014) or fear for their child's life (Bally et al., 2018), in addition to maintaining other everyday responsibilities, such as caring for the child's siblings. The findings of the present study therefore suggest that parents of children with rare CFAs **may** experience stress reactions that potentially have a significant and long-term impact on their psychological and physical health and relationships, as is also suggested in the broader paediatric literature (Woolf-King et al., 2017; Ljungman et al., 2014).

Risk Factors for the Development of Traumatic Stress Reactions

Evidence regarding risk factors for the development of traumatic stress reactions comes from reviews investigating this topic in general paediatric settings (Kassam-Adams et al., 2013; Pinquart, 2018). Research confirms that the subjective perception of threat is one of the major risk factors for the development of traumatic stress reactions, rather than "objective" markers of the severity of the medical condition, intensity of procedures, or other medical statistics (Kassam-Adams et al., 2013; Kazak et al., 2006; Balluffi et al., 2004). This is in line with the present findings, where the number of operations did not appear to be related to the degree of stress experienced. Hence, a treatment situation may appear uncomplicated to healthcare providers, but still be experienced as emotionally demanding by parents (Simpson & Catling, 2016). Assessments of stress reactions should therefore be based on the parents' personal perceptions of the situation, rather than the objective medical needs of the child. Trauma-informed care also highlights known psychological risk and/or protective factors, such as social support, previous life experiences, and pre-existing mental health difficulties as central (Price et al., 2015; Scrimin et al., 2009; Kazak et al., 2006). In the present study, participants had the opportunity to raise any issues they felt were important, but the interview guide did not specifically investigate known risk factors. Although this question may be best suited to quantitative research, the role of risk factors should be considered in future.

Positive Outcomes and Personal Growth

Recent research demonstrates a growing interest towards positive outcomes as a result of life-altering experiences (Kazak et al., 2005). In the current study, participants exemplified how parents, in spite of demanding experiences and potentially traumatic stress reactions, had experienced positive outcomes which they attributed to their child's condition and/or treatment-related experiences. As a result of their challenging

experiences, participants described themselves as more empathic, grateful, and accepting of other people's struggles. Participants also described a strengthened self-worth and felt they had become better parents. These findings mirror other research investigating post-traumatic growth and positive outcomes in parents of children with complex medical challenges (Bally et al. 2018; Ljungman et al., 2014), demonstrating how hope can be used to transform fear into a change in priorities, an enhanced appreciation of life, improved relationships with others, and personal strength. This is also in line with the theoretical description of traumatic growth as described by Tedeschi and Calhoun (1996).

The inclusion of positive outcomes in research and practice contributes to a more balanced view of psychological adjustment. Nonetheless, not all participants reported positive outcomes. It may be that some individuals possess a more optimistic outlook or construct positive outcomes as a way of reframing difficult experiences, while others struggle to articulate or identify positive outcomes in the context of their current life situation. While positive psychology seeks to redress the balance of psychological inquiry to also include personal strengths and growth, research and clinical experience of stress and trauma reminds us of the dialectic forces of suffering and growth that may often go hand in hand (Joseph & Linley, 2006).

Trauma-Informed Paediatric Care

In the wider health field, universal preventive interventions have been developed and include a wide array of evidence-based trauma-informed documentation for parents, patients and HPs. Examples are the National Child Traumatic Stress Network (www.nctsn.org), the Healthcare Toolbox (www.healthcaretoolbox.org), or the overview of resources available in the UK published in Sweeney et al. (2016). Improving HPs' understanding of stress reactions can promote identification and adequate management of symptoms in parents and children, thereby optimising health outcomes (Kassam-Adams et al., 2013). Trauma-informed care incorporates an evidence-based approach aimed at minimising the potential for treatment-related experiences to trigger or to serve as traumatic events by identifying distress, providing emotional support, encouraging coping strategies, and providing anticipatory guidance regarding treatment recovery (Marsac et al., 2016). The shared experiences within the current study suggest that trauma-informed care could also be helpful for parents of children with CFAs. Emotional stress reactions were clearly described by participants, yet few had accessed professional psychological support at the time of interview. Craniofacial teams are well positioned to identify and support

patients and their families who might be struggling to manage their stress reactions and refer those in need for assessments and follow-up as appropriate. Hence, systematic assessments of parental stress reactions may need to be provided in order to prevent the long-term impact of difficult treatment experiences.

Strengths and Limitations of the Current Study

The main strength of the present study is its relatively large sample, recruited from a pool of parents that are expected to be broadly representative of the Norwegian population. In-depth interviews facilitated the collection of individual experiences and allowed for investigation of associations between traumatic stress-inducing experiences and personal growth. Second, the interviews were performed by the same author, reducing the possibility for differences in interviewer technique and characteristics. Another strength was the inclusion of fathers in the study, since fathers' views and experiences are largely missing from the craniofacial literature (Klein et al., 2010).

Study limitations also need to be considered. First, traumatic stress symptoms were not systematically assessed. Stress reactions become problematic if they persist and disrupt parents' or children's everyday functioning, but the present study could not address to which extent stress reactions had become disruptive for the participants. Second, details from patient files regarding the number and timing of operations were not accessed, meaning that participants' experiences could not be interpreted in terms of more "objective" criteria. Although qualitative research is much needed within the present research field, quantitative studies will be necessary to validate the results of the present study, quantify the prevalence of emotional risk in the CFA population, and investigate mediating or predisposing factors. Third, participants who described positive outcomes may possess characteristics or coping strategies which allowed them to cope well at the time, rather than having experienced traumatic stress reactions, and therefore post-traumatic growth cannot be assumed. Nevertheless, growth following adversity does seem to be predictive of better emotional adjustment in the longer term, which may indicate that strengthening growth may be a useful interventional tool to help parents cope with adversity and stressful life events (Joseph & Linley, 2006). A fourth potential limitation is the impact of other diagnoses on parental stress reactions, such as developmental disorders and autism. The present study could not differentiate between the impact of a CFA and the potential additional impact of other conditions. The inclusion of CFAs coupled with additional conditions reflects a clinical reality that may need further attention (Lim et al., 2019).

Finally, the data collected in the present study relied upon participant recall of past events and narrative that has been constructed through participants' attempts to make sense of their experiences. Longitudinal research will be needed to understand the complexities and fluctuation of psychological adjustment to CFAs and the outcomes of CFA treatment. Despite these limitations, the findings of the present study suggest that parents of children with rare CFAs experience stress reactions that can have a significant and long-term impact on their psychological and physical health and relationships, as suggested in the broader paediatric literature (Woolf-King et al., 2017; Ljungman et al., 2014).

Conclusions

This study is one of the first to employ qualitative methods to explore the treatment-related experiences of parents of children with a rare CFA. Findings suggest that many parents experience stressful and potentially traumatic symptoms due to their child's complex medical treatment, which may in turn affect familial adjustment. The results of the present study highlight the importance of incorporating trauma-informed care in craniofacial treatment settings, in order to systematically identify and support those at risk of psychological distress. The study also sheds light on potential growth processes following difficult treatment experiences. Adverse life experiences are one way through which we may discover new strengths within ourselves or change our life's perspective (Joseph & Linley, 2006); a perspective that should receive more focus in future CFA research.

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