



Parental and health care professional views on psychosocial and educational outcomes in patients with cleft lip and/or cleft palate

Mia Stiernman^{1,2,3} · Kerstin Österlind¹ · Nichola Rumsey⁴ · Magnus Becker^{1,2} · Martin Persson⁵

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Abstract

Background Earlier research has investigated psychosocial and educational issues in populations of patients with cleft identifying several areas of concern. The objective of this study was to investigate current beliefs and knowledge about psychosocial and educational issues in parents and health care professionals (HCP) of children with cleft lip and/or palate (CL/P).

Method Parents were interviewed concerning the view of society on people with visible differences, their child's social life, educational progress, and information regarding psychosocial care. Interviews with HCPs concerned characteristics related with educational achievement, behavior, and social relationships. Fifteen parents of children 9 to 13 years of age with CL/P and 10 HCPs and were interviewed. Data from interviews was analyzed with thematic analysis.

Results Eight children were reported to have emotional issues related to their cleft. Eleven parents, however, did not perceive that their child was treated differently in society. HCPs expressed concerns regarding for example coping with being different, low self-esteem, shyness, disadvantage on first impression, and acceptance of themselves. A majority of the HCPs did not think patient cognition and behavioral or physical development were specifically affected.

Conclusions The results revealed that parent experience and views were diverse—from no specific problems related to the cleft, to both emotional and educational issues. The beliefs and level of knowledge in HCPs also varied. All HCPs, however, wished for more information and training regarding psychosocial issues.

Level of Evidence: Level VI, Diagnostic/Qualitative study

Keywords Cleft lip and/or cleft palate · Educational outcomes · Psychosocial outcomes · Interview · Qualitative results

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✉ Mia Stiernman
mia.stiernman@med.lu.se

¹ Department of Plastic and Reconstructive Surgery, Skåne University Hospital, Malmö, Sweden

² Institution for Clinical Sciences in Malmö, Lund University, Lund, Sweden

³ Plastikkirurgiska kliniken, Skånes Universitetssjukhus, Jan Waldenströms gata 18, Plan 2, 205 02 Malmö, Sweden

⁴ Department of Health and Social Sciences, University of the West of England, Bristol, UK

⁵ Department of Health and Society, Kristianstad University, Kristianstad, Sweden

Introduction

Previous studies have investigated psychosocial and educational issues in populations of patients with cleft identifying several areas of concern. Health care professionals working with patients with visible differences have reported that 30–60% of their patients experience psychosocial challenges. These professionals also wished to be able to address patient psychosocial issues but felt hindered in doing so by a lack of time, absence of an appropriate environment, and by the lack of appropriate knowledge and education [1, 2]. On the positive side, 1-day training courses have been demonstrated to have a significant positive effect on the confidence of health care professionals in dealing with psychosocial issues. Clarke and Cooper [1] reported that 66% of head and neck cancer nurses judged that participation in a 1-day course had significantly increased their skill set, strategies, and ideas for improving wellbeing in their patients. The remaining 33%

reported in the survey that the course had helped them “quite a lot” [1].

Results from research on the psychosocial impact of CL/P concerning social adjustment are inconsistent [3–6]; however, studies highlight a variety of psychosocial challenges associated with the condition. The psychosocial impact may not only affect patients during the active treatment phases of childhood and adolescence, but may also manifest as the patient grows older [7]. In a general health-related quality of life study, patients with unilateral cleft lip and palate aged 20–32 years reported negatively on social function, physical role function, and emotional role function when compared to patients aged 33–47 years. One reason for this difference could be the increased maturity and acceptance of their malformation over time [8]. A longitudinal study revealed that patients with cleft had a consistent pattern of internalizing behavior from 4 to 12 years of age. Girls have been reported to be particularly susceptible to very high levels of internalizing behavior during adolescence [9]. In contrast, another study showed that adolescents with a cleft did not demonstrate greater personality or adjustment problems than a control group without cleft [10].

In addition, patients with cleft have also reported a more positive social experience than the norm [11]. Theories that might explain these counterintuitive results include the possibility that these results could be the result of self-protective or coping processes based on denial. Patients could, on the other hand, be genuinely more accepting of themselves and more satisfied with their social functioning than other adolescents if, for example, their self-esteem was not as dependent on appearance than their counterparts who were unaffected by cleft [11]. A recent study showed no effect in the socio-economic status of patients with an oral cleft compared with an unaffected reference group. However, the subgroup males with CLP and CP had marginally lower incomes [12].

A Norwegian study addressed an additional potential influencing factor. In this study, 32% of patients with a cleft had known or suspected additional conditions. Psychosocial and/or cognitive functioning varied significantly when patients with additional conditions other than CL/P were included in the analysis, with results indicating that the population with a cleft and additional conditions had a higher level of psychosocial difficulties. Results from patients with CL/P only were, on the other hand, within the normal range for children without CL/P [13]. Opposing results and uncertainty within the research field may be due to the loose definitions applied to “other conditions” and the lack of reporting of additional diagnoses within the populations in the various studies. Another crucial factor contributing to the variation in results is the large variety of instruments and study designs used to investigate psychosocial issues, cognition, and educational achievement in the CL/P population and control groups [4, 14, 15].

Psychosocial aspects of patients with CL/P have been explored and reviewed in previous qualitative studies. [16–18] Issues identified as possible areas of concern by the patients themselves were as follows: regarding themselves as being different from others, low self-esteem, lack of recognition from others, appearance of the face, meeting new people, speaking in public, being understood, bullying in school, and passing the cleft on to future children. Parental psychosocial experiences have also been reviewed. [19] The reviewed qualitative data was of varying quality and the number of studies was few. Some of the psychosocial aspects of patients with CL/P were however reflected in the parents’ worries, such as acceptance of the child by peers, experienced teasing, finding a life partner, securing employment, and low self-confidence.

In relation to teasing and bullying, 75% patients reported they had been teased about their cleft features [20]. Also, 12% patients in a different study thought the teasing they had experienced was the most difficult cleft-related problem [21]. Negative effects on self-confidence as the result of a cleft were reported in two separate studies [20, 21]. Noor and Musa [20] reported this to be an issue for 83% of their sample with 73–91% of Turner et al.’s participants affected. Few patients informed the specialists at their cleft team about their concerns. Turner et al. [21], for example, reported that none of their participants had received help directly from their cleft team to cope with teasing.

As hypothesized by Topolski et al. [22], the lower scores on the environmental subscales of quality of life measures found in patients with facial differences could be due to the fact that they are often “picked on” because of their appearance [22]. Patients with a visible cleft were more likely to be teased than a non-cleft control population [23]. In a study of the relationship between satisfaction with appearance and harassment in 10 and 16 year olds, the presence of a visible difference was not a risk factor for reduced satisfaction with appearance when controlling for the patients’ subjective experiences of harassment. The author argues that such findings suggest that child perception of social experience is a primary factor in explaining child satisfaction with appearance—rather than the objective presence of a visible cleft [23].

Recent research highlights a potentially significant deficiency in educational level in people affected by cleft. Two studies reported that patients with CL/P had lower school grades and lower odds of achieving pass in mandatory education compared to norm population or a matched sample without a cleft [24, 25]. People affected by cleft also had lower levels of academic achievement compared to matched classmates even when controlling for several potential demographic and socioeconomic confounders [26]. They were also three times more likely to use special education services than children without CL/P [27]. A demographic analysis of Swedish adults with cleft and a population of non-cleft controls showed

a lower level of education in the cleft population [28]. A recent study showed that males with CLP and females with CP had a marginally reduced rate of completion of intermediate level education [12]. One study, however, found no significant differences in academic achievement between children with clefts and their non-affected siblings [29].

Concerning cognitive issues, a meta-analysis study from 2012, revealed only one difference in cognitive function in people affected by cleft compared with the norm. This difference was that those with a cleft had significantly impaired language skills. Statistically significant impairments in other domains of cognition were also found, but the data did not meet the criteria for study homogeneity and publication bias [15]. Two large Scandinavian population-based studies found that there was a higher risk of being diagnosed with a psychiatric disorder in nonsyndromic patients with clefts compared with individuals without clefts [30, 31] and compared with the patients' siblings [31].

Differences in IQ between cleft diagnoses have also been found. These results are, however, not unanimous in their findings. Two population-based studies investigating the intelligence of 18-year-old Swedish and Norwegian men revealed firstly that Swedish men with cleft palate only (CPO) had a lower level of general intellectual capacity than the norm population. However, the group of patients with cleft lip with or without cleft palate (CL±P) did not score significantly differently from the norm however [32]. In the second study, Norwegian men with CP had lower IQ than men with no birth defect. The scores for men with CL/P however did not differ with the comparison group after matching for birth order, maternal education, maternal age, and marital status [33]. An American study on male adults with CL/P reported scores within the normal IQ range, but lower than non-CL/P controls when matched for age, sex, and parental socioeconomic status. In this study, participants with CPO scored higher than those patients with CL/P [34].

Possible reasons for differences in IQ or academic achievement have been discussed in previous publications [3, 14, 33–38]. Examples of “primary” and “secondary” explanatory factors exist. Craniofacial and cerebral development could be potential “primary” causes. Hearing loss, speech deficits, and social effects as a result of altered appearance are, on the other hand, examples of issues thought to cause “secondary” IQ or grade deficiencies. In a study of the cerebral structure of children with CL/P, magnetic resonance imaging (MRI) revealed that CL/P patients had smaller intracranial volume, cerebral volume, cerebellar volume, and less brain tissue [38]. Statistically significant differences in cerebral morphology between patients with CL/P and a non-cleft control population were revealed. Cerebral morphology differences in those affected by a cleft also correlated with IQ [39]. A further study employing the same methodology by the same author correlated aberrant cerebral morphology with social dysfunction

[40]. These findings support the notion that differences in those affected by cleft are the result of primary causes, impacting cognition and social behavior. Thus, interdependency between “primary” and “secondary” causes is likely to contribute to a combined final effect.

Previous research has reported children with CL/P to be at higher risk for developmental difficulties than the general population [13]. Findings of low-level academic achievement from a young age in children with a cleft underpin recommendations for the routine early screening of children with oral clefts [13, 14, 26, 35–37]. As previously stated, this screening would ideally involve collaborations between schools and craniofacial teams [26].

Although it is difficult to extrapolate with confidence on the basis of the conflicting finding of research to date, psychosocial and educational issues are a vital ingredient of positive wellbeing [41] and in particular, to the psychological health of people with visible differences. Seventy-one percent of an outpatient plastic surgery population with a variety of visible anomalies expressed the desire for access to an appropriately trained professional to support them in their efforts to deal positively with their appearance-related concerns [2]. In the absence of research focussing specifically on the perceived relevance of wellbeing and academic outcomes in those affected by clefts, the objective of this study was to investigate current beliefs and knowledge of health care professionals (HCPs) and parents of children with CL/P in southern Sweden regarding psychosocial and educational issues related to growing up with a visible cleft.

Materials and methods

This study formed part of the EU funded Erasmus + project 2015-1-SE01-KA202-012291 IHEM—*Improving the long-term outcomes in children with congenital anomalies by implementing an Innovative Health Educational Module for staff in health care and NGO settings* [42]. The aim of the study was to inform the development of a training package for HCPs relating to the psychological and educational issues associated with congenital anomalies and visual differences. As the study was exploratory, a qualitative, semi-structured interview methodology was employed. The interview templates for HCPs and parents were developed on the basis of the literature reviewed above by co-authors NR and MP. Research ethics board approval and informed consent from participating parents and health care professionals were obtained.

The catchment area of this study was the southern part of Sweden. All children born with CL/P in this area are treated at the center. Approximately 40 new cases are registered each year. Treatment protocol includes primary lip closure at 3 months of age, hard and soft palate closure at 12 months

of age, and bone transplant to alveolar clefts at approx. 8 years of age. Velopharyngeal flap surgery, secondary cleft surgery, therapy with speech and language pathologist, and placement of grommets are carried out when necessary. The social counselor also provides counseling to the children and families. However, the educational progress and psychosocial health of children and young people treated in this clinic are not routinely followed or the focus for screening by any staff at the center. Although work is underway to implement a routine patient-reported outcome measure, at the time of writing, child and parent reported outcomes are not yet routinely collected.

Parents of patients with CL/P

Fifteen parents of children with a visible cleft were interviewed. Inclusion criteria were that the child had a visible cleft, was 9 to 13 years of age, and has no syndrome related to the cleft. For more details of the diagnosis according to Allori et al. [43] and treatment of the children, see Table 1. The sample comprised nine mothers and six fathers, 37 to 59 years of age, all of whom were members of two consecutive samples of patients visiting the cleft center for a routine appointment with their child and thought to represent typical cases. Each parent was approached by the author, MS, and invited to participate. Thirteen interviews were conducted by telephone and two interviews were conducted face to face. Interviews took approximately 30 min and were conducted individually with each participating parent. The cleft center's social

counselor, KÖ, conducted the interviews. The interviews were tape recorded and/or captured using an interview template. Interviews continued until the interviewer, KÖ, and the author, MS, concluded that a saturation effect had been achieved with no further themes brought up compared to earlier data. [44]

Parents were asked about their views about how society responds to people with visible differences, about their child's social life, educational progress, and on their confidence in tackling any concerns. In addition, they were also invited to discuss whether or not they felt they had sufficient support and information regarding such issues and if not, their views about how such information and support should be provided.

Health care professionals

Ten interviews were conducted face to face with HCPs by the author, MS. Interviews with the HCPs took approximately 40 min each. Participants were cleft specialists with a variety of professional backgrounds, together forming a targeted sample. All were working directly with patients with CL/P and other diagnoses associated with visible differences (CATCH 22, Tessier, Van der Woude, Apart, Stickler, hemangiomas, burn accidents, congenital facial paralysis, hemifacial microsomia, Treacher-Collins, Down syndrome, Pierre-Robin, microtia, micrognathia, and ptosis). Their experience with this patient group varied between 4 and 40 years, (mean 19 years). Two surgeons, two nurses, two assistant nurses, two orthodontists, and two speech and language pathologists

Table 1 Description of participant's children's age, sex, diagnosis, and treatment

Patient ID	Age	Sex	Cleft diagnosis	Age at lip adhesion (months)	Age at lip repair (months)	Age at palate repair (months)	Age at alveolus repair (years)	Age at surgical treatment for VPI (years)	Speech therapy	Hearing disorder
1	10	Girl	bCL + P	*	*	*	8 and 9	–	Yes	Yes
2	13	Girl	uCL + P	3	9	18	9	–	No	No
3	9	Girl	bCL + P	3	9	12	9, 10, and 11	4	Yes	No
4	9	Girl	uCL ± A	–	5	–	9	–	No	No
5	9	Girl	uCL + P	–	*	16	8 and 9	–	No	No
6	10	Boy	uCL ± A	–	6	–	10	–	No	No
7	10	Boy	bCL + P	3	11	17	8 and 10	–		No
8	11	Boy	uCL + P	3	9	14	9	11	No	Yes
9	13	Boy	uCL + P	4	9	19	7 and 10	5	Yes	Yes
10	10	Boy	uCL + P	–	4	12	10	–	No	No
11	13	Boy	uCL + P	**	**	**	10 and 11	4	Yes	No
12	10	Boy	uCL + P	–	3	11	8 and 9	5	Yes	No
13	13	Boy	uCL ± A	–	3	–	7	–	No	No
14	13	Boy	bCL + P	**	**	25	7	–	Yes	No
15	9	Girl	uCL ± A	–	5	–	9	–	No	No

*Adopted, lip repair approx. 1 year of age

**Adopted, lip and palate repair before 3 years of age

participated. The interview included questions regarding participants' views of the key psychosocial and educational issues related to growing up with a cleft. HCPs were interviewed concerning characteristics that they believed to be related to positive educational achievement, behavior, and social relationships. They were also asked if they would like to have more information and training regarding psychosocial issues associated with CL/P. Answers were recorded on a specifically designed interview template.

Data analysis

Data from this narrative style qualitative study was analyzed with thematic analysis. [45] In this process, common themes and subthemes in the narratives of all interviews are identified and presented to illustrate a wide set of experiences related to the studied phenomenon. Quotes are presented with respective themes to illustrate the parents' experience. Themes and quotes were checked with five of the parents who participated in the study and whose quotes were used.

Results

Parents of patients with CL/P

The main themes to emerge from thematic analysis were *Experiences related to societal views on visible differences*, *Parental concerns related to their child's cleft*, *Social relationships*, *Educational progress*, *Emotions and behavior*, and *Information and support from cleft team*. Themes and subthemes are presented in Fig. 1.

Experiences related to societal views on visible differences

Eleven parents expressed the view that society did not treat their child differently due to their visible difference and did not think that their child had suffered any negative impact.

Nobody treats him differently because of the cleft. As a person yes, but not because he has a cleft.

Five reported that relatives and people outside the family pitied their child, asked questions, or commented about the cleft and that some people looked or stared in a disturbing way. Some parents had also prepared their child to cope with the reactions of other people to their appearance.

You don't see the person behind, only the scars and the flat nose.

We have prepared him quite early that he probably has to be prepared that people will react. And especially that those who react negatively will somewhere use it against him.

All parents but one felt that there was nothing particularly positive about his or her child having a visible difference. One parent stressed that their child had received extra support in school because of her hearing deficiency.

We live in a shallow world. And everything that is different, unknown, you are afraid of. Parental concerns related to their child's cleft

Most parents had concerns regarding the future of their child. Some of this concern, however, was regarded as "normal parental concern."

But it is, all parents have different troubles... (laughter).

Eight parents had cleft-specific concerns related to speech development, being understood, dental development, recurrent ear infections, asymmetry of the face, inability to correct the visible difference, concerns over failed cleft surgery, and bullying because of the cleft. One mother was worried that her child was beginning to feel sad. The child had asked why she had been born with a cleft and was having difficulties coping. There were, however, seven parents who had no concerns about the future of their children. Some emphasized that their child had strengths that would help them overcome any disadvantage of a visible difference.

[He] is very social. He is good at looking out for himself.

Social relationships

All parents reported that their child had friends in school and in addition, most children had friends outside of school and/or online. Nonetheless, 13 parents reported that their child was, or had been, teased in school or received unwanted attention related to the cleft and the visible difference.

My worry is that there will be more [bullying] the older he gets. Of course when they are younger they have another tolerance toward each other, toward a visible difference. (...) You can also feel worry about when he will go into the teenage years and it becomes messy regardless.

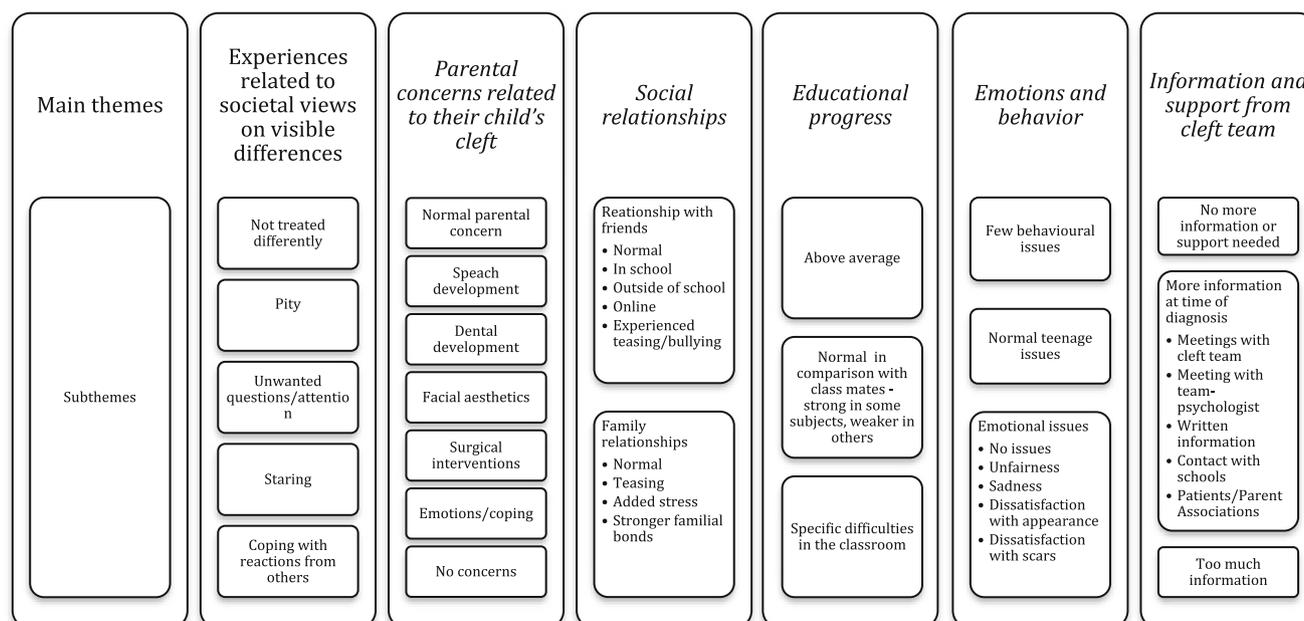


Fig. 1 Main themes and subthemes identified in interviews with parents of patients with CL/P about their views on psychosocial and educational outcomes. The variability of the subthemes reflects the parent's variability of views

Even if they only come up because they have noticed, or ask questions, or point or something he calls it bullying. Then I say that that is not bullying, that curiosity, but he feels like he is being attacked.

One parent noted that discomfort in class decreased once the child had got to know the new school class. Another parent noted that she thought her child had benefitted from being in the same group of children since a very young age.

Fourteen parents reported good relationships within the family. In two cases, the child was teased within the family or had expressed that he or she was different and not loved as much as his or her siblings. Most families experienced additional stress and loss of income in relation to hospital visits and operations. Two parents experienced stress at time of diagnosis.

It was only when he was very very little that it was a thing. Then, you know, it isn't anything we think about. He doesn't get any special treatment because of it [the cleft].

Two parents hypothesized that the family relations had become stronger because of the child with a cleft. Because of the visible difference and the challenge of coping with the cleft and treatment other, less important, problems were put in perspective and not allowed to take as much energy.

The thing is that these types of challenges, or whatever you call it, when you have a child with a special need, make you... have different kinds of reflections or relations. Eh, like, you take life differently maybe. Eh, you see things in a more sober way maybe. Little things are not, eh, you don't need to fight about small unimportant things. You can concentrate on things that are important.

Educational progress

Regarding educational progress, 11 parents reported that their children were similar when compared with their peers. Four parents said that their child achieved above average in school.

I think she is just like her friends [in school].
Eh, it varies a bit. He is very good in some subjects. And in other subjects he could spend some more time.

Six parents noted, when asked further, that their child had specific difficulties in the classroom such as concentration, dyslexia, withdrawnness, or hyperactivity. All the six parents who reported that their child had specific difficulties also reported that their children achieved above average or similarly to their peers in school.

Emotions and behavior

Parents reported very few issues in relation to the behavior of their child. Only one child was reported to have behavioral issues, but had no clear diagnosis of these difficulties had been made. Nine children had emotional issues related to their appearance and for eight, this related to their cleft. Parents of three children reported that the child thought it was unfair that they had been born with a cleft. Three children thought they looked ugly and one was worried that the scars would not improve and would always be visible. Seven children were not bothered by their cleft and were satisfied with their appearance.

It has been very hard. (...) It has to do with his appearance (...) That he doesn't talk about it, but his perception is that he is so ugly.

He has had periods when he has been sad that he has a cleft lip and palate. Eh, but it has not at all been to the extent that I was worried about when he was little.

Information and support from cleft team

Parent reports were varied when discussing support and information about psychosocial issues associated with cleft and visible differences. Answers ranged from not needing any more information or support, to feeling that they would have appreciated more information at time of diagnosis.

I think it varies a lot depending on the person. I have a group on Facebook for clefts that I follow sporadically. (...) I notice there that there are many who are pregnant and expecting child with a cleft who ask questions and who are very worried and don't know at all. How, eh... So you notice that there is an unmet need for network and information.

In two cases, parents thought that they had too much information at an early stage. One father who had received the cleft diagnosis at a prenatal ultrasound and was given information about the condition at a prenatal cleft consultation reacted with relief when the child was actually born and was not as sick or negatively affected as he had feared.

And then he is born – and what's the problem?

Those who wanted more information suggested it should be delivered in the form of meetings with the entire cleft team

together with other families, or alternatively, in other forums, such as a patient association.

I think it could be healthy for them to get to meet and see other children with this type... So that they can ask questions and air their thoughts. I think this could be important for these children. And also to strengthen the self-confidence further. It surely isn't the same for all the other children like it is for my boy, there are surely those who feel very sad because of this.

Two parents also thought that the school should be informed about the cleft diagnosis to enable them to tackle potential teasing more effectively. One mother stressed the importance of the active involvement of a psychologist in the team. She also felt that it was important that all staff in the team should be able to respond to her child's appearance-related emotional issues instead of dismissing the issue.

We are governed by appearance whether we admit it or not. And it is very important that we admit that to our children. That this is important, and that 'no, you are just as pretty as everybody else' is just ignoring people's problems. (...) If there isn't a psychologist then I would at least like to have a written document about different... how to respond to your child with a visible difference – what do you say, what to do, how to manage. At school, at home, emotionally, everything. Because I have felt really lost there.

One parent preferred written information since it can be difficult to remember all the information relayed at a meeting at the hospital.

The best would be to have a written document, and if you had any questions there would be someone available to call.

Health care professionals

The major themes to emerge from the thematic analysis were *Views on visible differences in society*, *Impact of cleft on social functioning*, *Impact of cleft on psychological functioning*, *Impact of cleft on educational performance*, *Factors related to positive adjustment/resilience*, and *Further training needed for health care professionals*. Themes and subthemes are presented in Fig. 2.

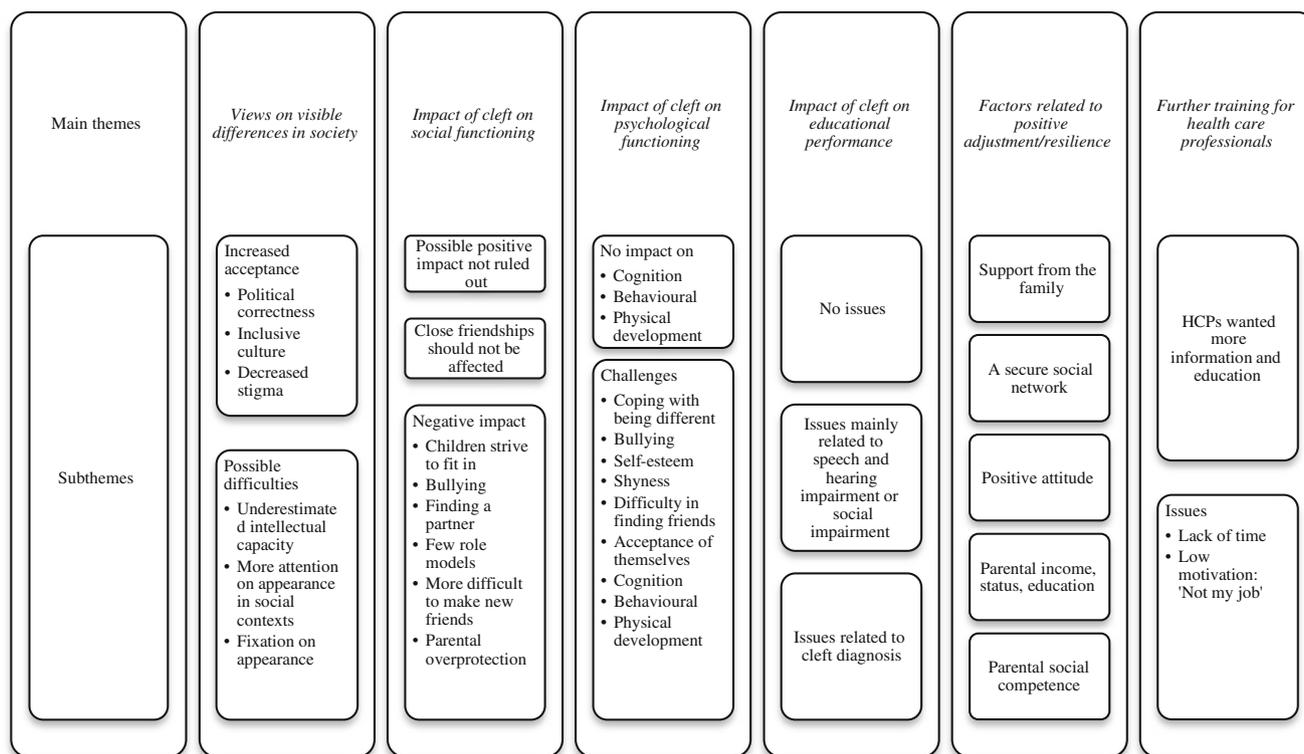


Fig. 2 Main themes and subthemes identified in interviews with health care professionals about their views on psychosocial and educational outcomes. The variability of the subthemes reflects the health care professionals' variability of views

Views on visible differences in society

The health care professionals reflected on two characteristics in society; on the one hand, political correctness and an inclusive culture increase acceptance and decrease stigma associated with visible differences in schools and workplaces. On the other hand, participants commented that even if people affected by cleft are broadly accepted in society, their intellectual capacity might be underestimated due to their visible difference. Those who are not familiar with the diagnosis may also be more reserved toward people with a visible difference causing social isolation. Also, as more attention is being placed upon appearance in social contexts, people are becoming increasingly fixated on appearance, people with an unusual appearance are perceived to be at a disadvantage.

Impact of cleft on social functioning

HCPs reported no positive impacts of having a visible difference. One HCP did not rule out that there might be a few positive impacts, but could not pinpoint what these might be. Participants did, however, believe there could be a negative impact and expressed the view that children affected by cleft have to strive to fit in with their peers. Concerns about bullying in childhood and adolescence, finding a partner, and the lack of positive role models with visible differences were

mentioned. Some HCPs believed that the amount of impact depended on whether or not the child had good support from his or her family or not. Others correlated the social impact with the degree of visible difference.

All HCPs agreed that the social functioning of children with CL/P was adversely affected by a visible difference. Regarding social relationships, answers were more diverse. Six HCPs argued that “real friends” or close friendships and family relationships should not be affected by having a visible difference. Another comment was that it could be more difficult to make new friends. One HCP believed that a cleft could negatively affect close relationships, for example, as the result of parental overprotection.

Impact of cleft on psychological functioning

A variety of psychosocial challenges for this patients population were identified, including coping with being different, striving to fit in, bullying, finding an atmosphere in which they are accepted, low self-esteem, shyness, difficulty in finding friends, being at a disadvantage in the process of the first impressions formed of them by others, avoidance of situations where they feel afraid of failing and finally, acceptance of themselves, dissatisfaction with appearance. Many of these challenges were believed to depend on the reactions the patient experienced from their surrounding social, cultural, and family environment. A majority, seven HCPs, did not think

patient cognition and behavioral or physical development were affected in any specific way.

Impact of cleft on educational performance

HCPs' views on the impact of cleft on education were also varied. Three HCPs did not associate cleft with any educational issues at all and pointed out that having a cleft was not in itself a problem in the academic domain. Three HCPs believed that the association between the cleft and possible educational issues was a consequence of impairments to speech and hearing impairment or to challenges to social functioning, rather than result of the actual cleft. A third group of four HCPs proposed that the cleft diagnosis was more directly associated with educational issues because of, for example, differing brain morphology.

Factors related to positive adjustment/resilience

Factors identified by HCPs as contributing to higher levels of resilience and positive psychosocial outcomes in their patients were support from the family, a secure social network, a positive attitude, and the child's personality in general. Two HCPs believed that income or parental education did not have any impact on educational or psychosocial outcomes. Eight HCPs, on the other hand, correlated parental income, social status, and educational outcomes to levels of engagement in treatment and also to more favorable outcomes for the patient. One HCP expressed the hope that financial and social variables were not predictive of patients' outcomes, but nevertheless thought that this might be the case. One view given was that a child with a visible difference might not fit in with an otherwise "perfect" family. Parental social competence was considered to be of higher importance than income or educational level by three HCPs.

Further training for health care professionals

None of the HCPs had received any formal training regarding the psychological factors and processes involved in adjustment to cleft. The source of information was seminars and conferences. All HCPs agreed that they would like more information and education regarding appropriate treatment and pathways of referral for CL/P patients with psychosocial issues. It was however noted by some HCPs that there was a lack of time to address such issues in their clinic, that they did not want to stigmatize the patients by asking about their psychosocial health or suggesting treatment and neither did they feel that this was "their job." Most HCPs felt that they could alert the social counselor in the team if they believed that their patients could benefit psychosocial counseling.

Discussion

The responses from the HCPs taking part in this study revealed a diversity of views on most topics. Opinions about the potential associations between educational achievement and CL/P were especially diverse, as were the respondents' thought on the characteristics connected with favorable outcomes in educational settings, behavior, and social relationships. Some of the answers from HCPs may be less negative or pessimistic due to lack of knowledge of possible complications of cleft, for example, in relation to language skills [15]. This, combined with the fact that none of the HCPs interviewed had received any formal training in psychosocial health in this patient group, highlights the need for continuous professional development and training in this specialized field. The need for all members in a cleft team to be aware of common psychological challenges and to possess the skills necessary to meet the needs of patients experiencing psychosocial difficulties is crucial. Parents, too, highlighted the importance of addressing this important aspect of care.

Reviews on interventions for both children and adults targeting psychosocial health with cognitive behavioral therapy and social skills training provide some, but weak, evidence of their efficiency [46–48]. It is summarized that interventions are important to people with psychosocial issues related to their visible differences, but methodological weakness in studies investigating interventions only provides weak support their effectiveness [46]. In spite of the this previous research, few HCPs appeared to be aware of the potential of counseling or psychological treatment for patients with cleft to improve psychological wellbeing, to alleviate the negative impacts of shyness, or to improve levels of self-confidence. This lack of awareness could in turn lead to under-utilization of existing pathways of referral for psychological treatment. It may also reflect an unwillingness to stigmatize patients by suggesting they should be referred for further treatment. This fear among professionals has been commented on in earlier research. [48] This review concluded, however, that patients seem to accept treatment targeting social skills training, not finding the treatment stigmatizing.

HCPs reflected on a number of possible negative outcomes related to growing up with a cleft such as underestimation by others of the intellectual capacity of the affected person, difficulty fitting in, bullying, or finding a partner. Eleven out of 15 parents, however, did not perceive that their child was treated differently in society. This mismatch in the perceptions of HCPs and parents is striking and HCPs should be alerted to the need to identify and respond to every patient's individual experience. [49] Meanwhile, eight of 15 children were reported to have emotional issues due to their cleft and parent reports

showed that specific difficulties experienced by children were primarily due to the reaction of others to their child's appearance. This finding supports earlier research. [16, 17, 20, 22, 23] Earlier research also highlights the fact that patients rarely raise these issues during consultation with cleft teams.

Six parents initially reported that their children achieved higher or similar results in school compared with peers despite also mentioning specific problems with mathematics, reading, or concentration. This contradictory information from parents stresses the need for a careful assessment of the educational experience and outcomes for each child. In these cases, important information and issues of clinical relevance to the cleft team were only unveiled after further discussion on issues other than general performance in school. The high proportion of emotional issues and the fact that six out of 15 parents reported that their children had specific difficulties in the classroom also stresses the need for routine early screening. The need for clear referral pathways for cognitive assessment and counseling or psychological treatment for cleft patients is also clear.

Limitations of the study

This qualitative study is based on interviews with HCPs and parents from only one cleft center. Although this is an exploratory study to guide future research and the content of training, caution is necessary when making assumptions about other centers where different routines for psychosocial screening and treatment are implemented. Major themes are however in line with earlier research and shed light on issues regarding the variability present in the subjective views of individual participants, and the importance of including a focus on the psychosocial health and educational progress of patients as an integral part of care [50]. Interviews with most parents of children with a visible cleft were conducted by telephone. Non-verbal channels of communication were therefore limited. Participants' responses may have been more nuanced had face-to-face interviews been used in all cases. Interviews with HCPs were not tape-recorded and quotes could therefore not be extracted to illustrate the themes.

Conclusion

In conclusion, this study investigated current views of parents and health care professionals on psychosocial and education issues in children with CL/P. The results revealed that parental experience and views were diverse, ranging from the opinion that there were no specific

problems related to the cleft—to a clear articulation of both emotional and educational issues. The views and level of knowledge of HCPs also varied. All HCPs, however, wished for more information and training regarding psychosocial and educational issues and treatment. This study also shows that important information and issues regarding emotional wellbeing and educational achievement were only unveiled after focused discussion on these issues. Time and space for such discussion needs to be planned within routine follow-up.

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Compliance with ethical standards

Conflict of interest The authors, M. Stiernman, K. Österlind, N. Rumsey, M. Becker, and M. Persson, have no conflicts of interest.

Ethical statement

Ethical approval was obtained from Ethical Board in Lund, Sweden, reference no. 2016/1024.

Informed consent Informed consent was obtained from all participants.

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