SOCIAL-EMOTIONAL COPING IN ADOLESCENTS WITH CRANIOFACIAL ANOMALIES

By

Rany Bous, B.D.S.

Submitted in partial fulfillment of the requirements for

the degree of Master of Science in Dentistry

Thesis Advisor: Manish Valiathan, BDS, MDS, DDS, MSD

Case Western Reserve University School of Dental Medicine

Department of Orthodontics

June 10, 2018

DEDICATION

To my parents, who taught me the pleasures found in hard work, the appreciation for learning, acceptance of people's differences and to develop a passion for life. Their substantial support along with their immense love for life have always inspired me.

ABSTRACT

Background: Numerous studies have investigated psychosocial adjustment levels among adolescents with cleft lip and/or palate, however, our understanding of other craniofacial conditions remains to be scarce. The primary purpose of the current study is to bridge some of the gaps in the literature through comparing psychosocial adjustments among three groups of adolescents: craniofacial conditions other than cleft lip and/or palate (CFA), adolescent with cleft lip and/or palate (CLP), and a control group (CON). Secondary objectives are identifying social factors that may influence adjustments levels, and comparing informant agreements among parent, teacher and self-reports.

Design: Cross-sectional, multi-informant, controlled survey study.

Participants: 11-17 year olds. 49 with CFA other than CLP, 42 with isolated CL/P, and 55 controls. Total = 146.

Outcome measures: The Strengths and Difficulties Questionnaire (self-report, parent report, and teacher report).

Results: Parent reports showed significant differences among the 3 groups, with CFA exhibiting significantly higher total difficulty scores than CLP (p=0.027) or CON (p=<0.001), and CLP significantly higher than controls (p=0.016). Subscales showed that CFA had higher peer problems compared to CLP (p=0.001), and CLP was higher than CON (p=0.05). CFA group displayed a significantly higher frequency of abnormal

psychosocial adjustment (34.7%), compared to 17% among CLP group, and only 3.8% for controls.

Informant agreements were moderate, yet significant, for parent reports vs self-reports, and parent reports vs teacher reports. However, correlations between self-reports vs teacher reports were not significant. The most predictive factor for was the frequency of being bullied, followed by having a good friend. However, the number of parents in the household or the number of siblings were not significant. The four studied social factors combined explained 44.2% of the variance.

Conclusions: A general trend was observed where CFA group displayed the highest psychosocial difficulties, followed by CLP, and the lowest were the CON group. Within the subscales, peer problems constituted the biggest challenge that faces both CFA and CLP groups. Social factors, especially being bullied or having a good friend, could significantly mediate and influence the psychosocial adjustment levels.

KEY WORDS: cleft lip and palate, craniofacial conditions, psychosocial adjustment, informant agreement, SDQ.

TABLE OF CONTENTS

Page

DEDICATION (optional)…….. ii

ABSTRACT ……. …….. iii

REVIEW OF THE LITERATURE ……. .1

LIT REVIEW REFERENCES (ALPHABETICAL BY AUTHOR) ……. 30

STATEMENT OF THE THESIS ……. 43

MANUSCRIPT FOR PUBLICATION ……. 47

MANUSCRIPT REFERENCES (FORMAT ACCORDING TO JOURNAL) ……. .83

APPENDICES (all data collected) ……. 93

1. **Table 1.** Demographics……..……..93
2. **Table 2.** Parent reports (n=142) - SDQ scores and subscales……94
3. **Table 3.** Self reports (n=144) - SDQ scores and subscales…….……95
4. **Table 4.** Teacher reports (n=86) - SDQ scores and subscales……..96
5. **Figure 1.** Mean SDQ scores for all groups. ……97
6. **Table 5.** Frequencies of normal, borderline, and abnormal SDQ scores…….98
7. **Figure 2.** Categorical distributions among the groups……99
8. **Table 6.** Informant agreement among parent, teacher, and self-reports……..100
9. **Table 7.** Regression analysis for the predictive factors……..101
10. **Figure 3.** Effect of significant social factors on total difficulty score…….101
11. **Figure 4.** Frequencies of being bullied, and frequencies of having a good friend across the groups……..102
12. **Figure 5.** Sample size calculation. ……..102
13. **Figure 6**. Parent version of the SDQ. ……,,,,,,,,.103
14. **Figure 7.** Self-report version of the SDQ.……104
15. **Figure 8.** Teacher version of the SDQ.……105
16. **Figure 9.** The parent version impact supplement of the SDQ…….....106
17. **Figure 10.** Impact supplement for the self-report version of the SDQ…..…..107
18. **Figure 11.** Impact supplement for the teacher version of the SDQ…..……...108
19. **Figure 12.** Scoring the SDQ……..……..109
20. IRB APPPROVAL.....112

21. INFORMED CONSENT……113
22. CHILD ASSENT FORM……..……...115
23. Teacher Recruitment Letter……..……....117
24. **Table 8.** Comparison between our CON and US normative data……...119

1-HISTORICAL PERSPECTIVES

Centuries ago, humans did not possess our current medical knowledge of embryology and development, and thus, they interpreted congenital deformities based upon myth, religion and superstition.[1] Infants with congenital deformities were presumed to harbor evil spirits, or were regarded as a punishment from the gods, to the extent that some cultures believed they were a result of women copulating with the devil.[2] Such beliefs have led many ancient cultures to the tragic acts of abandonment, exposure, or killing of newborns affected with such anomalies.[3] Shockingly, even some of the greatest philosophers of all time advocated these practices. In his *Politics*, Aristotle wrote "*let there be a law that no deformed child shall live.*"[4] Likewise, Plato, in the *Republic*, advocated that these practices benefit the society by eliminating evil spirits.

Our attitudes towards the etiology of these congenital conditions started to shift with the introduction of modern science during the Renaissance. In 1651, William Harvey presented his theory of teratogenesis, where he reasoned that the presentation of clefts is just an extension of a "normal" state observed in embryos. The works of Gregor Mandell in the 19th century have revolutionized our understanding of genetics, and allowed us to appreciate the genetic nature of congenital deformities, instead of attributing them to myth and legends.[5] With that understanding, some cultures started abandoning their superstitious beliefs and adopting more scientific approaches.

Currently, the majority of the Western world has banned such harsh practices. However, some cultures and African tribes still hold their beliefs about the supernatural etiology of congenital deformities.[6] For generations, they have ignored modern medicine, and resorted to traditional healers and cleansing rituals to treat their children and rid them of what they presumed to be evil spirits.[7] Whereas in China, it is still common to hear stories about such acts of abandonment of children with congenital deformities.[8] Such interactions can be explained by the innate basic human responses, surprise, or uncertainty regarding how to behave, rather than cruelty or barbarism. Ryan et al. concluded that such aversion and prejudice towards facial disfigurement could be explained by the fact that humans evolved to avoid individuals with signs of disease, even when they know that their condition is non-contagious.[9]

While the superstitious beliefs and rituals have diminished to a large extent, especially in Western cultures, our basic human instincts remain to tarnish our societal attitudes and acceptance of the affected individuals.[10,11]

2- CRANIOFACIAL ANOMALIES (CFA)

With the advent of modern medicine, our understanding of the biological nature of congenital deformities has broadened, and we have come to an understanding of the complex embryological basis behind their development. Today, the term "craniofacial anomalies" (CFA) refers to a diverse group of congenital deformities that affect the head and the facial structures.[12] These conditions exhibit a wide range of severities and prevalence rates, with cleft lip and/or palate (CLP) being the most common at about 1 in 700 birth in the United States.[13] Other craniofacial anomalies include, but are not limited to, various forms of craniosynostosis, hemifacial microsomia, cleidocranial dysplasia, fetal alcohol syndrome, Pierre Robin sequence, Treacher Collins syndrome and many other syndromes. They are described as "*anomalies*", because they deviate from what is considered to be normal.

Being born with a craniofacial condition can have various compounding impacts on the affected persons and their families. In addition to the evident facial disfigurement, the effects of such rare conditions may constitute—but are not restricted to—cognitive impairment, speech problems, hearing loss, abnormal development, and psychosocial disturbances. Hence, the affected individuals frequently require multiple surgeries and medical interventions throughout the years, creating an enormous burden of care.

The medical advancements have tremendously progressed to explain the scientific causes of these deformities, and the reconstruction procedures have become

ever more sophisticated, in efforts to not just restore the functional aspects of the affected individuals, but to also normalize their appearance. All in hopes of improving their quality of life and psychosocial experiences.

However, the societal attitudes towards people with craniofacial anomalies have remained largely static, burdening them with the stigma that accompanies their deformities throughout their lives, struggling to integrate themselves in a society that values appearances more than ever before.

3- STIGMA

Erving Goffman, one of the most influential modern sociologists, defined stigma as "*The phenomenon whereby an individual with an attribute which is deeply discredited by his/her society is rejected as a result of the attribute. Stigma is a process by which the reaction of others spoils normal identity.*"[14] Goffman categorized stigmas into 3 main categories: Stigmas associated with physical deformities, stigmas associated with personality traits, and stigmas associated with ethnicity or religion.[14]

The association between appearance and social stereotyping is established in social science research.[15] Teachers were found to perceive more attractive children as more intelligent and socially successful, than less attractive children.[16,17] Macgregor considered facial disfigurement to be a "social disability" because of its impact on not just the individual, but also their social interactions.[18] Thus, stigmas associated with physical deformities are of major concern to individuals with craniofacial conditions, who are already dealing with an enormous burden of care due to their conditions.

Tragically, despite the advancements in our knowledge of these conditions, the social stigmas remain to affect our perceptions of this vulnerable population, reflecting adversely on the psychosocial well-being and quality of life of the affected individuals.[19,20,21] This is complicated further by the negative self-perception of the

affected person, leading to feelings of dissatisfaction, worthlessness and social isolation.[20,21,22]

Kesande et al. found that, in Uganda, 40% of mothers of children with clefts believed that children with clefts are regarded as outcasts in their community.[23] When Adeyemo et al. surveyed these perceptions in Nigeria, they found that 73% percent of the mothers were ashamed of having a child with a cleft, and 25% wished the baby was never born, whilst 20% of the surveyed friends and relatives refused to hold the affected baby.[24] Such beliefs and responses may not be as extreme in more developed countries, however, the stigmas still exist even if represented in subtler forms, and extend throughout life. In 2012, Pausch found that 57% of the participants in his study use animal terms to label clefts, which was deemed hurtful by cleft patients.[25] Moreover, Krueckeberg et al. found that preschoolers with craniofacial anomalies were rated as significantly less attractive and less friendly than their non-affected peers.[26] Teachers observed that children with cleft lip and palate display greater social inhibitions and lower achievement scores, when compared to non-affected children.[27] Richman noted that although children with cleft lip and palate have normal IQ scores, there is a tendency for them to be underachievers, which is hypothesized to be a result of the behavioral inhibitions, and the lower expectations by parents and teachers, as a self-fulfilling prophecy.[28] Kapp-Simon concluded that children with clefts have a lower self-concept than their non-affected peers, as they perceived themselves to have lower social skills and to be more often

sad and angry than their peers.[29] As early as 1939, Baker noted that individuals with facial disfigurements are more likely to attribute their negative social interactions to their stigmas, which may reflect onto their psychosocial functioning.[30] Examining the social interactions of adolescents with CFA revealed that they were more marginalized and less likely to be involved in conversations, when compared to their non-affected peers.[31] Research has shown that individuals with facial differences encounter daily social challenges, including stares, comments, and questions about their facial differences.[32] Such stigmas can also be depicted in the harsh forms of teasing or bullying, especially during childhood or adolescence. Studies have shown that 60%-75% of children with CFA are regularly teased, which is significantly higher than the ranges for the general population in the United States, which ranges between 9% and 50%.[33,34,35,36,37,38] Modecki et al. conducted a meta-analysis of 80 studies analyzing the prevalence of bullying among teenagers, and reported a mean prevalence rate of 35%, which is significantly lower than the prevalence among adolescents with CFA.[39]

Unfortunately, the adverse effects of victimization can extend throughout life, and were found to elevate the risk of psychopathology, depression and suicide among the general population.[40] The same conclusions have been established for the CFA population, as Feragen et al. have concluded that experiences of teasing among adolescents with CFA can lead to a lower self-esteem, decreased satisfaction with appearance and elevated risk of depression.[35,40] A gender factor has been observed as

well, where twice as many females reported teasing than males, and the impact of teasing over time was greater for adolescent females than for males.[35]

The implications may extend into adulthood, as adults with CFA are less likely to be married or employed, more likely to receive disability pension, reported less social support, and lower engagement in social activities.[41,42] McWilliams reported that cleft patients are more likely to drop out of high-school, and less likely to be married, when compared to their non-affected siblings.[43] Affected individuals have also reported that they found it difficult to initiate romantic relationships, which can reflect on their quality of life.[44] These stigmas may extend to the level of discrimination, where they can have everyday implications. Sarwer et al. concluded that 38% of adults with craniofacial anomalies reported being discriminated against in employment or social settings.[45] In an attempt to understand these discriminations during job interviews, Madera et al. found that interviewers who evaluated applicants with facial deformities focused more on the deformity, remembered less facts about the applicant, and rated the facially stigmatized applicants lower than the non-affected ones, demonstrating that such discriminations may affect employment chances of the affected individuals.[46]

Rumsey et al. revealed that even children exhibit stereotyping behaviors against individuals with facial deformities.[47] When asked to rate pictures of adults with facial deformities before and after surgery, 75% of 11-year olds perceived the pictures of adults before surgery to be less friendly, less helpful, and more fearful, compared to their after surgery pictures.[47] Therefore, it is of no surprise that adults

with facial differences are concerned about the challenges of living in a modern society that struggles to accept people with differences, while prioritizing appearance more than ever before.[48]

Such stigmas and negative social experiences can be associated with subjective negative self-perceptions and lower levels of psychosocial adjustment. Berger and Dalton found that social experiences can predict the extent of psychosocial adjustment among adolescents.[49] In a similar vein, Feragen et al. established an association between social experiences and subjective self-evaluation, suggesting that positive social experiences are associated with better emotional adjustment.[50]

The aforementioned social challenges that face this vulnerable population have led many investigators to suspect that individuals with craniofacial conditions may be at a higher risk of psychosocial impairment, and hence gained a vast interest among researchers.

4- PSYCHOSOCIAL IMPLICATIONS AND QUALITY OF LIFE

Understanding the impact of craniofacial conditions on the quality of life and psychosocial well-being of the affected individuals is crucial for craniofacial teams, in order to provide a holistic approach to treatment. In the 1980s, researchers found that hospital settings lacked counseling services for CLP patients, who seemed to be struggling with social maladjustments.[51,52] Likewise, Broder concluded that 56% of individuals with CFA had problems warranting psychological referral.[53] As our knowledge of the potential psychosocial impacts of these conditions evolved, it has become the standard of care for craniofacial teams to employ psychological and social services to support the affected individuals and their families.[54,55] Even though such services are highly valuable to the affected individuals and their families, the increased risks of psychosocial maladjustment remains to affect this population.

A major psychological risk factor for adolescents with craniofacial conditions is a poor body image, which refers to a person's own perceptions of their body.[56] Individuals with facial differences can often feel dissatisfied with their appearance, and even after multiple surgical interventions, many parents and patients uphold concerns about their appearance and speech.[57,58] Marcusson et al. found that almost

50% of adults with repaired clefts were dissatisfied with their appearance and wished to have more operations.[59] With gender adjustments, Feragen et al. showed that adolescent girls with a visible cleft were at a higher risk for being discontent with their appearance than boys.[40] Moreover, they concluded that among individuals with CFA, peer harassment was related to dissatisfaction with appearance.

Since body image and satisfaction with appearance is an integral part of self-esteem, a negative body image can reflect adversely on the person's self-worth and quality of life.[60,61] Marcusson et al. concluded that dissatisfaction with facial appearance was the most significant predictor of depression.[59] Interestingly, recent studies concluded that the subjective evaluations of the severity of the condition demonstrate higher correlations to the adverse psychosocial outcomes, while objective measures of the severity showed low correlations.[62,63,64] Multiple studies have shown that subjects with CFA or CLP tend to have a lower self-concept when compared to non-affected individuals.[21,44,65,66] A recent study evaluated the body self-esteem of Australian CLP patients across three age groups and concluded that adolescents and adults have a significantly lower self-esteem scores when compared to normative data.[67] In one study, 73% of CLP patients reported that their confidence had been adversely affected by their condition.[33] Maris et al. found that children with hemifacial microsomia were concerned about their appearance, and demonstrated lower self-esteem compared to their non-affected twins.[68]

Several studies explored behavioral problems among children with CFA, and although the findings have been inconsistent, it may be appropriate to presume that

children with CFA are at a higher risk of developing behavior problems. Teachers reported poorer behavior ratings for children with CFA, compared to non-affected peers.[69] Likewise, parents' reports showed that behavioral problems among young people with non-syndromic craniosynostosis were significantly higher than the general population.[70] An analysis of children with trigonocephaly showed that 20% of the children needed an assistant in the classroom due to behavioral challenges due to an elevated frequency of developmental, educational, and behavioral problems.[71] Statistically higher rates of behavioral problems were also reported by parents of children with syndromic forms of craniosynostoses.[72,73] When adjusting for gender among individuals with Apert syndrome and Muenke syndrome, boys displayed significantly higher frequencies of behavioral problems than girls.[74]

A longitudinal follow-up study found that 31% of children with various CFA exhibited behavior problem scores in the clinical range by either teacher or parent reports.[75] Speltz et al. found similar results in a sample of five-to-seven years old children with CFA.[76] Interestingly, they found that girls with CFA displayed more externalizing or types of behavior problems, than either boys with CFA or children without CFA.[76] In contrast, Cloonan et al. found no significant differences between the frequency of behavioral problems among children with non-syndromic craniosynostosis compared to controls.[77] Similarly, parents of children with hemifacial microsomia reported similar levels of behavioral difficulties to those reported by matched controls.[78]

In regards to quality of life (QOL), many investigators have reported that individuals with facial differences are at a higher risk of having lower QOL scores compared to non-affected ones.[79,80,81] Wehby et al. surveyed craniofacial health professionals to rate the health related QOL of patients with CLP, and found a modest decrease in their QOL.[80] In a similar vein, Damiano et al. concluded that the QOL declined significantly with the occurrence of severe speech problems, and that older children with CLP had lower scores compared to younger children.[82] Another study concluded that children with facial differences had lower QOL in comparison to children with no chronic conditions.[81] However, their quality of life was similar to that of children with other chronic disorders.[81] Teachers reported that in comparison to controls, children with Hemifacial Microsomia have higher frequencies of internalizing problems, lower social skills and less peer recognition.[74] Similar results were found among adults with CLP, when compared to controls.[59] Marcusson et al. found that adults with CLP rated their quality of life significantly lower than controls in the areas of life-meaning, family life, and private economy.[83] However, they had significantly higher scores regarding ability to make the most of leisure time and to be active.[83]

In contrast, Wogelius et al. found no significant differences in QOL between Danish children with CLP and controls, while Oosterkamp et al. found no significant differences between adults with bilateral CLP and unaffected adults from the Netherlands.[84,85] Aravena et al. reported similar findings for children and teens with CLP.[86] Likewise, other investigators concluded that parent reports and self-reports

have shown normal QOL in children with a wide range of CFA.[87,88] Other studies that focused on adults with specific craniofacial conditions, such as Cherubism, Apert, and Crouzon syndromes found satisfactory QOL in comparison to normative scores.[89,90,91]

Identifying the impact of CFA on psychological adjustment is a crucial factor in understanding the complexity of such disorders. Psychological or emotional adjustment, is defined as the ability to maintain emotional equilibrium in the face of internal and external stressors.[92] Evidently, individuals with CFA and their families face numerous stressors throughout life. Hence, it is understandable that many researchers have investigated the psychological adjustments among this population. In a classic systematic review, Endriga and Kapp-Simon deduced that although research suggests that many children with craniofacial conditions do not experience psychological problems, it is evident that significantly more children with CFA would be expected to demonstrate psychological, behavioral, and emotional difficulties that are of clinical concern.[93] Singh and Moss reported a significant psychological impact of various CFA among Nepalese patients, although they found no gender effects.[94] In the same vein, a recent meta-analysis analyzed the psychosocial impact of CLP and found that that men with CLP are at a higher risk of developing psychosocial problems compared to women, and the impact was higher for adults compared to adolescents.[95] Moreover, they concluded that irrespective of age, gender, or culture, individuals with CLP have lower psychosocial functioning than non-affected individuals.[95] This was supported by another recent systematic review by Nicholls et

al, where they reported that 89% of the 28 papers that were included in the review indicated that having an orofacial cleft leads to an elevated risk of developing psychosocial difficulties for children.[96]

A longitudinal study with a robust sample of 6,462 Danish individuals with clefts showed that patients with cleft palate had an elevated risk of being hospitalized due to mental disorders.[97] Moreover, they concluded that patients with CLP had an increased risk of substance abuse, but the risk for schizophrenia or bipolar disorder was not statistically significantly different from comparison groups.[97] Maris et al. compared children with Hemifacial Microsomia to their non-affected twins, and reported that the affected twins displayed a heightened risk for developing psychological difficulties.[68] Parents and teachers of children with CLP reported various significantly more psychosocial problems among their children.[98,99] Demir et al. analyzed the prevalence of specific psychological disorders among CLP patients and found an increased risk of developing psychopathologies, especially social anxiety disorder and major depressive disorder, among children with clefts.[100] In their sample, 45% of children with CLP were diagnosed with anxiety disorders, while 30% showed symptoms of depression.[100] Similarly, Ramstad et al. reported that anxiety, depression, and palpitations were twice as prevalent among CLP compared with controls.[101] Likewise, Nicholls et al. found similar results amongst an Australian cohort of adults with CLP, where they stated that 78% of participants reported experiencing at least one psychosocial health issue and more than half experienced anxiety and/or depression.[102] However, they displayed equivalent education levels,

annual incomes, employment rates, and home ownership levels to the general population.[102] In comparison to the vast number of studies focusing on psychological adjustments of individuals CLP, those that focus on CFA other than CLP are relatively scarce.[103] Tovetjärn et al. stated that 73.9% of adults with Apert syndrome reported periods of depressive mood, compared to 23.3% in the comparison group.[104] Another study found lower scores of emotional well-being among children with syndromic craniosynostosis, as rated by parents.[73] Likewise, 33.3% of young adults with Crouzon syndrome reported periods of depression compared to 19.1% in a matched control group.[41]

Several investigators found associations between psychological adjustment and social experiences. Parents considered children who had been teased to have increased prevalence of psychosocial problems.[98] Roberts found positive correlations between higher rates of discrimination and elevated levels of depression and anxiety in adults with CFA.[105] A similar association was found between emotional problems and to limitations in social activities.[39] In the same manner, Roberts and Mathias revealed that social support, self-esteem, and fear of negative evaluation could predict the incidence of anxiety among individuals with CFA, while depression was predicted by lower self-esteem and reduced social support.[106] Feragen et al. investigated the effects of developing romantic relationships on adolescents with CLP, and showed that significantly fewer adolescents with CLP reported being in a romantic relationship compared to controls, and significantly more adolescents with CLP reported having never been in a relationship.[107] However, in that study, depressive

symptoms and self-esteem among teenagers with CLP were not found to be associated with difficulties achieving romantic relationships.[107]

Two particularly alarming studies investigated the mortality and suicide rates of adults with CLP.[108, 109] Christensen et al. followed a cohort of Danish adults with CLP for a total of 170,421 person years, and showed that CLP is associated with an increase in mortality rate, not just in infancy, but even up to age 55.[108] In the same vein, Herskind et al. reported that the suicide rate among Danish adults with clefts was double that of the non-affected population.[109]

In contrast, other studies have found levels of adjustment to be in line with comparison groups. Kapp-Simon et al. evaluated the psychosocial adjustments of adolescents with CFA, and found that their self-perception scores and social skills are within the normal range.[110] Another study conveyed that the psychosocial adjustments of children with CLP and their mothers were not significantly different from that of the normal population.[111] Roberts and Mathias reported that mean scores of depression and anxiety among adults with CFA were within the normal range.[41] Similar results were reported by parents of children with complex craniosynostoses, where they found no significant differences between affected individuals and comparison groups in relation to emotional functioning.[73] Interestingly, Feragen et al. showed that adolescents with CLP reported higher scores of emotional wellbeing and global self-worth, compared to the control group.[107] Another study of adolescents with facial differences showed that the affected individuals reported less depressive symptoms than the comparison group, which may be attributed to increased

emotional resilience.[50] Through semi-structured interviews, Patel and Ross showed that many adults with CLP reported that despite of the social challenges, they were satisfied with their educational attainments, communicative abilities, family life and marital life.[112]

The impact of the psychosocial implications of CFA may extend to the families of affected individuals as well.[93] The initial parental reactions to the birth of a child with CFA are often feelings of shock, grief, anger, or anxiety.[113,114] This is often followed by gradual adaptation to the child's condition and an improvement in the emotional response.[113] In addition, some parents describe feelings of guilt or blame themselves for their child's condition.[115] Some studies revealed that parents may feel socially stigmatized as well. Mothers have reported increased sensitivity to the reactions of the friends and perceiving feelings of repulsion in public.[116,117] One study has shown that parents of children with craniosynostosis reported a lower quality of life, mostly impacting the psychosocial aspects of their lives.[70] Kramer et al. studied the impact on 130 families having children with clefts, and reported a generalized modest impact on the families'quality of life.[118] The strongest effects were on coping strategies and personal aspects, while the financial impacts and the effect on siblings were the lowest.[118] Likewise, parents of children with CFA experienced significantly higher levels of parenting stress compared to controls.[119] Thamilselvan et al. found that behavioral problems among children with CLP can impact the quality of life of the parents, and positively correlated to parental depression, stress, and various aspects of QOL.[120] Reciprocally, increased parental distress may lead to poorer

psychosocial adjustment levels among children with CFA.[119] Bannink et al. examined the effects on QOL of children with syndromic craniosynostosis and their families, and noted an adverse effect on family activities.[72] Moreover, they concluded that syndromic craniosynostosis has a significant adverse impact on the health-related QOL of these children and their parents.[72] Crerand et al. found that 32% of the families of children with CFA reported at least one risk factor and 7.8% displayed multiple risks.[121] Another study showed that even siblings of children with CFA may have an elevated risk of developing psychosocial disturbances.[122] Conversely, Baker et al. reported that parents of children with CLP may exhibit high levels of positive adjustment and social support.[123]

Overall, it is safe to conclude that several studies suggest the presence of adverse effects on the psychosocial well-being, various aspects of the QOL, and long-term health of the affected individuals and their families. However, these findings have been inconsistent, a number of studies reported psychosocial functioning of the affected individuals to be in-line with, or even better than, comparison groups. The variations in the findings may be attributed to some specific challenges that face this particular field of research, which will be discussed in a latter chapter.

5- STRENGTHS AND COPING MECHANISMS

On a superficial level, it may seem appropriate to conclude that individuals with CFA face an elevated risk of developing psychosocial problems, but it is evident that many of the affected individuals exhibit levels of psychosocial functioning in-line with, or even better, than comparison groups. This has driven more recent studies to shift their focus towards positive outcomes and understanding the coping mechanisms that may help individuals cope better with their conditions. Studies have shown that social skills are a major predictor of adjustment levels.[49,93] Kapp-Simon et al. reported that social skills training is effective in improving the frequency of social interactions among adolescents with CFA, and thus may enhance their psychosocial adjustments.[124] Likewise, increased support from friends and family was found to correlate with better adjustment levels.[50,123] Berger and Dalton evaluated various coping mechanisms, and concluded that individuals exhibiting

problem-focused styles of coping were better adjusted than those who used avoidant styles of coping.[49] In an thought-provoking narrative, Strauss proposed three theories to guide us into accepting the "blessing in disguise" associated with CFA.[125] First, he suggests that clinicians should create an optimistic and positive environment around the time of birth and diagnosis. Second, he proposed that clinicians should start asking the patients and their families about their strengths and positive qualities. The third proposal is launching a new social science model built around understanding resilience and strengths of this population, rather than the difficulties.[125] Several adults with CFA believed that meeting other people with similar conditions could be valuable in providing support, and leading to better coping, while others stated that receiving professional psychological support had been very beneficial.[126] Lowenstein et al. stated that volunteering or helping others appeared to be a prominent coping strategy among individuals with facial differences.[127] Moreover, some individuals reported other coping mechanisms such as family support, faith, spirituality, journaling, being involved in activities and sports, or compensating for their differences by doing well in school.[127,128] Kapp-Simon et al. found social skills and athletic competence to be the greatest predictors of adjustment, accounting for 73.5% of the variance within their sample.[81] In contrast, some individuals seem to adopt unhealthy coping mechanisms, such as alcohol abuse.[127]

In an attempt to explore the strengths of this population, qualitative studies revealed that individuals living with visible differences could reach higher levels of satisfaction through placing greater value to other aspects of their lives, such as personality, friendships and family, rather than focusing on appearances. [128,129,130] Some individuals revealed that they can even feel fortunate, when they compare their lives to others with more severe conditions.[126] People with facial differences have reported attracting good friends and romantic partners, who value their personalities more than their appearance. [126,130] Some adults with CLP have reported that having a facial difference made them unique in a positive way, while others believed that it enabled them to grow into better people. [130,131] In one study of adults with CLP, one participant said "It's made me a better person. I'm a lot more understanding, I wouldn't judge anybody… I wouldn't change it for anything."[131] Several authors highlighted the elevated levels of resilience among this population, where coping with a facial difference has helped them develop resilience to face various life challenges.[50,130,131] Parents have also stated the strengths of their affected children, such as perseverance, willpower, and elevated social skills.[132] In addition, parents have reported the positive influences of having an affected child, such as teaching the family lessons about accepting differences, elevating their spirituality, increasing their involvement in community services, and even improving their relationships.[128,130]

A remarkable narrative by Robert Kelton, who himself is affected with a craniofacial condition, describes his journey of coping with his condition, and the factors that influenced him the most.[133] He discussed how the dedication and support he received from his family and healthcare team had a positive impact on his life.[133] Kelton wrote: *"I have no doubt that much of my striving and much of my determination to succeed stem directly from the adversities inherent in craniofacial anomalies"*, describing how the adversity itself has promoted his success.[133] His recommendations to healthcare professionals included encouraging patients to seek out others with similar conditions to help them learn from each other, and to maintain a sense of humor and optimism.[133] In a qualitative and quantitative study involving adults with CFA and parents of children with CFA, Eiserman explored the positive outcomes and found that the majority of individuals endorsed their condition as an integral part of who they are.[130] He revealed that half of his sample would not choose to remove the experience of facial difference from their lives or their children's lives, if given the opportunity.[130] Moreover, 45% of parents reported the experience to be equally positive and negative for their children, and 81% of affected adults reported that it was more positive than negative.[130] In response to one of the interview questions, a participant said: *"Having a cleft lip and palate has helped create much beauty inside me and I definitely honor who I've become because of it"*.[130]

As evident from the current body of knowledge, the variability in responses, diversity of coping mechanisms, and the complexity of the of CFA makes it challenging to draw definite conclusions regarding their psychosocial implications.

\

6- CHALLENGES FACING CRANIOFACIAL PSYCHOSOCIAL RESEARCH

Systematic reviews have attributed the inconsistency of findings in this field of research to the variations in study designs, populations, inclusion criteria and the wide variety of instruments used for assessing various aspects of adjustment. [103,134,135] A common challenge for most studies is the small sample size of cases, and the variety among the craniofacial conditions, and within each condition, when mixed up in one sample, which can confound the results. Such limitations can be attributed to the rarity of such conditions, and thus, some authors suggested conducting multi-center studies, to help overcome these limitations. [103,135]

In addition, the variability of informants whose perspectives are measured, which includes self-evaluations, parent, teacher, or health professional reports may have led to the variation in the findings, especially due to the objective nature of psychometric evaluations.[103,134,135] Informant discrepancies have been highlighted in the literature, where different informants can report dissimilar evaluations of the psychosocial adjustment levels of the affected individuals.[33] Children and parents displayed poor levels of agreement, regarding satisfaction with appearance, and psychosocial evaluations.[33,111,136,127] Berger and Dalton found that mothers of children with CLP reported elevated levels of difficulties, although the children's self-evaluations were within normal levels.[111] They portrayed various explanations for such discrepancies, such as increased levels of stress or sensitivity among mothers, or adolescents under-reporting as a form of self-protection.[111] In contrast, other studies found that self-reports of children with CLP indicated lower levels of adjustment than parent reports.[137,138] This was attributed to various reasons, such as increased vulnerability of affected children, increased tolerance threshold among parents, or the fact that parents may lack insight into the child's social life, as they do not observe them throughout the entire day.[137,139] Another explanation is that parents reports tend to be derived from a long-term perspective, while children may report symptoms of more recent incidents at the time of evaluation.[137] However, discrepancies between parent and self-reports is not specific to individuals with CFA, as reference groups exhibit similar levels of poor agreement.[139] It is important to note that poor levels of agreement do not necessarily suggest that the reports are invalid, but rather reflect

unique perceptions of each informant.[137,140] To overcome this drawback, researchers emphasized the need for more studies that include multiple informants besides self-reports, such as parents, teachers, and clinicians.[103,137]

Hunt voiced his concern about the fact that the literature tends to report the psychosocial functioning of CLP children in a general way, which may conceal specific problems these individuals may have.[134] Other methodological limitations found in the majority of the studies are the lack of appropriate control groups, and the lack of longitudinal studies.[103,134] Longitudinal studies are especially important in understanding the directionality of associations, and the complex impacts of such multifaceted conditions, especially as these effects may shift or develop over time.[103,134] Interestingly, Feragen et al. discussed the overall lack of qualitative studies, such as structured interviews.[103] Qualitative studies may not provide us with numbers and means, but they allow the affected individuals to voice their own unique thoughts and perspectives, which can be especially valuable in helping researchers gain an insight into what it is like to live with a facial difference.[103] Another gap lies in the scarcity of studies that focus on CFA other than orofacial clefts. Although numerous studies have investigated the impacts of clefts, the research on other CFA remains to be limited.[103] In addition, Hunt et al. discussed the lack of studies that focus on why some children with CLP may cope better than others, and highlighted the need for more studies that explore coping mechanisms, and the strengths rather than the difficulties.[134]

7-THE STRENGTHS AND DIFFICULTIES QUESTIONNAIRE (SDQ)

Due to the wide variety of instruments that can be used to measure various aspects of the psychosocial adjustments of individuals with CFA, researchers have pointed out the need for more uniform studies, utilizing similar instruments, in order to be able to perform meta –analyses and compile homogenous data that may allow us to draw better conclusions. However, it is extremely challenging for an investigator to decide which instrument would fit this population the most.

In 1997, Dr. Robert Goodman developed the Strengths and difficulties questionnaire (SDQ), as a method of improving and updating the long established Rutter parent questionnaire.[141] The SDQ is a brief screening tool for psychosocial difficulties and positive strengths in children and adolescents that exists in various versions. Since then, it has been translated to over 80 languages, and has been mentioned in over 4,000 publications from 95 countries. All versions of the SDQ ask about 25 attributes, some positive and others negative. Each item is scored ‘‘not true,’’ ‘‘somewhat true,’’ or ‘‘certainly true’’ (score range, 0 to 2).[141] These 25 items are divided equally into 5 subscales to assess each of five distinct aspects of behavior: 1) emotional symptoms, 2) conduct problems, 3) hyperactivity, 4) peer problems, and 5) prosocial behavior. Each subscale includes five items that are positively or negatively worded. The first four subscales are added together to generate a total difficulties score (20 items with a score range of 0 to 40), while the last subscale addresses the strengths (five items with a score range of 0-10).[141]

In addition, extended versions have an impact supplement which identifies the psychological impact on different aspects of the adolescent’s life, and/or follow-up questions that can be used for longitudinal studies.[142]

Although SDQ scores can be used as continuous variables, it is sometimes convenient to categorize scores. Goodman categorized the scores of the SDQ scores into three bands, ‘normal’, ‘borderline’ and ‘abnormal’. These bandings were defined founded on a population-based survey in the United Kingdom, attempting to choose cut-points such that 80% of children scored ‘normal’, 10% ‘borderline’ and 10% ‘abnormal’.[141]

Another strength of the SDQ, is that is provides different versions for multiple informants, including parents, teachers, and self-reports, with corresponding items and scales. This allows researchers to obtain various perspectives from multiple informants. A multi-informant approach has been recommended when investigating psychosocial issues in children and adolescents, in order to provide a comprehensive assessment of their psychosocial adjustment levels.[143,144,145]

The SDQ has shown satisfactory validity as it was found to be highly correlated to the Child Behavior Checklist and the Rutter Questionnaire.[141,146] In correlation to semi-structured interviews, the SDQ was significantly better than the Child Behavior Checklist at detecting inattention and hyperactivity, and at least as good at detecting internalizing and externalizing problems.[146] However, the SDQ is only around a fifth of the length of the Child Behavior Checklist, and thus may be preferred by participants due to its brevity.[146] In a robust study that included a sample of over ten-thousand 5 to 15 year olds, the SDQ exhibited satisfactory internal reliability for both parent and self-reports as well, which adds to its strengths.[147] Moreover, the SDQ has shown high specificity and moderate sensitivity in predicting child psychiatric diagnosis, making it a valuable screening instrument.[148,149,150]

Because of the aforementioned advantages, coupled with its simplicity and brevity, it is of no surprise that the SDQ has been increasingly adopted by many researchers and clinicians since its development. Vostanis regarded the SDQ as one of the most extensively used instruments in children psychosocial research across different cultures.[151] In craniofacial research, the SDQ has recently been utilized by a

number of investigators. [49,111,137,138] In addition, the Oslo craniofacial team has adopted the SDQ as their instrument of choice for routine psychological assessment of children with CFA. [138,152] However, we are unaware of any studies in the craniofacial field that have utilized the extended impact supplement of the SDQ.[126]

Therefore, we concluded that it is appropriate to consider the SDQ as one of the most appropriate tools to study the psychosocial adjustments for adolescents with CFA, and thereby the decision to use it for our current study.

REFERENCES

1. Converse JM, Hogan VM, McCarthy JG. Cleft lip and palate. In: Converse JM, editor. Reconstructive Plastic Surgery. 2nd ed. Philadelphia: Saunders; 1977. p. 1930.

2. Savona-Ventura C. The influence of the Roman Catholic Church on Midwifery Practice in Malta. Medical History 1995;39:25-6

3. Dorrance GM. The operative story of cleft palate. Philadelphia: Saunders; 1933.

4. Dunn PM (2006). "Aristotle (384–322 bc): philosopher and scientist of ancient Greece". *Archives of Disease in Childhood: Fetal and Neonatal Edition*. 91: F75–7.

5. Sturtevant AH. A history of genetics. New York;Harper & Row,1965;9-32

6. Olasoji HO, Ugboko VI, Arotiba GT. Cultural and religious components in Nigerian parents' perceptions of the aetiology of cleft lip and palate: implications for treatment and rehabilitation. The British journal of oral & maxillofacial surgery. 2007; 45: 302-305,

7. Dagher D, Ross E. Approaches of South African traditional healers regarding the treatment of cleft lip and palate. The Cleft palate-craniofacial J, 2004; 41:461-469.

8. Johnson KA. China's Hidden Children: Abandonment, Adoption, and the Human Costs of the One-Child Policy. Chicago, IL: University of Chicago Press; 2016.

9. Ryan S., Oaten M., Stevenson R.J., Case T.I. Facial disfigurement is treated like an infectious disease. Evolution and Human Behavior, Vol. 33(6): 639 – 64

10. Pausch, N. C., Winter, K., Halama, D., Wirtz, C., Yildirim, V., & Nattapong, S. Psychosocial acceptance of cleft patients: Has something changed? Journal of Oral and Maxillofacial Surgery, 2006 ;20: 19–26.

11. Partridge, J. Psycho-social reflections on craniofacial morphogenesis.Seminars in Cell and Developmental Biology, 2010;21: 333–338.

12. Buchanan, E. P., Xue, A. S., & Hollier, L. H., Jr. (2014). Craniofacial syndromes. Plasticand Reconstructive Surgery, 2014; 134, 128e–153e.

13. Tanaka SA, Mahabir RC, Jupiter DC, Menezes JM. Updating the epidemiology of isolated cleft palate. Plast Reconstr Surg. 2013;131: 650e–652e

14. Goffman, E. Stigma: Notes On The Management Of Spoiled Identity. Englewood Cliffs, N.J.: Prentice-Hall. 1963.

15. Langlois, J. H., Kalakanis, L., Rubenstein, A. J., Larson, A., Hallam, M., & Smoot, M. Maxims or myths of beauty? A meta-analytic and theoretical review. Psychological *Bulletin, 2000;126*(3), 390-423.

16. Clifford, M.M. and Walster, E. The Effect of Physical Attractiveness on Teacher Expectations, Sociology of Education, 1973;46(2):248–258

17. Martinek T. Physical Attractiveness: Effects on Teacher Expectations and Dyadic Interactions in Elementary Age Children. Journal of Sport Psychology 1981 3:3, 196-205

18. Macgregor, F. After plastic surgery: Adaptation and adjustment. New York: Praeger. 1979

19. Kapp K. Self-concept of the cleft lip and or palate child. Cleft Palate J. 1979;16(2):171-6.

20. Topolski TD, Edwards TC, Patrick DL. Quality of life: how do adolescents with facial differences compare with other adolescents. Cleft Palate Craniofac J. 2005;42(1):25-32.

21. Broder H, Strauss RP. Self-concept of early primary school age children with visible or invisible defects. Cleft Palate J. 1989;26(2):114-7.

22. Macgregor FC. Some psychological problems associated with facial deformity. AM Social Rev 1951; 16:629-638.

23. Kesande T, Muwazi LM, Bataringaya A, Rwenyonyi CM. Prevalence, pattern and perceptions of cleft lip and cleft palate among children born in two hospitals in Kisoro District, Uganda. BMC Oral Health. 2014; 18:14-104.

24. Adeyemo WL, James O1, Butali A. Cleft lip and palate: Parental experiences of stigma, discrimination, and social/structural inequalities. Ann Maxillofac Surg. 2016;6(2):195-203.

25. Pausch NC, Herzberg PY, Wirtz C, Hemprich A, Dhanuthai K, Hierl T, Pitak-Arnnop P. German animal terms for oral cleft deformity: a Leipzig survey. J Craniomaxillofac Surg, 2012 40:236–242.

26. Krueckeberg SM, Kapp-Simon KA, Ribordy SC. Social skills of preschoolers with and without craniofacial anomalies. Cleft Palate Craniofac J. 1993;30(5):475-81.

27. Richman LC. Behavior and achievement of cleft palate children. Cleft Palate J. 1976 Jan;13:4-10.

28. Richman LC, Eliason M. Psychological characteristics of children with cleft lip and palate: intellectual, achievement, behavioral and personality variables. Cleft Palate J. 1982;19(4):249-57.

29. Kapp-Simon K. Self-concept of primary-school-age children with cleft lip, cleft palate, or both. Cleft Palate J. 1986;23(1):24-7.

30. Baker WY, Smith LH. Facial disfigurement and personality. JAMA 1939; 112:301-304.

31. Kapp-Simon KA, McGuire DE. Observed social interaction patterns in adolescents with and without craniofacial conditions. Cleft Palate Craniofac J, 1997 34:380–384

32. N. Rumsey. Body image and congenital conditions with visible differences. T.F. Cash, T. Pruzinsky (Eds.), Body image. A handbook of theory, research, and clinical practice, The Guilford Press, New York, USA (2002), pp. 226-233.

33. Turner, S., Thomas, P., Dowell, T., Rumsey, N., & Sandy, J. Psychological outcomes amongst cleft patients and their families. British Journal of Plastic Surgery, 1997; 50, 1– 9.

34. Lorot-Marchand A., Guerreschi P., Pellerin P., Martinot V., Gbaguidi CC., Neiva C., Devauchelle B., Frochisse C., Poli-Merol ML., Francois-Fiquet C. Frequency and socio-psychological impact of taunting in school-age patients with cleft lip-palate surgical repair. Int J Pediatr Otorhinolaryngol. 2015;79(7):1041-8.

35. Feragen KB, Stock NM. A longitudinal study of 340 young people with or without a visible difference: The impact of teasing on self-perceptions of appearance and depressive symptoms. Body Image. 2016; 16:133-42.

36. Craig W, Harel-Fisch Y, Fogel-Grinvald H, Dostaler S, Hetland J, Simons-Morton B, Molcho M, de Mato MG, Overpeck M, Due P, Pickett W. A cross-national profile of bullying and victimization among adolescents in 40 countries. Int J Public Health. 2009; 54 Suppl 2:216-24.

37. Nansel TR, Overpeck M, Pilla RS, Ruan WJ, Simons-Morton B, Scheidt P. Bullying behaviors among US youth: prevalence and association with psychosocial adjustment. JAMA. 2001;285(16):2094-100.

38. Brunstein Klomek A, Marrocco F, Kleinman M, Schonfeld IS, Gould MS. Bullying, depression, and suicidality in adolescents. J Am Acad Child Adolesc Psychiatry. 2007; 46(1):40-9.

39. Modecki KL, Minchin J, Harbaugh AG, Guerra NG, Runions KC. Bullying prevalence across contexts: a meta-analysis measuring cyber and traditional bullying. J Adolesc Health. 2014;55(5):602-11.

40. Feragen KB, Borge AI. Peer harassment and satisfaction with appearance in children with and without a facial difference. Body Image. 2010;7(2):97-105.

41. Roberts RM, Mathias JL. Psychosocial functioning in adults with congenital craniofacial conditions. Cleft Palate Craniofac J. 2012;49(3):276-85.

42. Danino A, Gradell J, Malka G, Moutel G, Hervé C, Rosilio C. Social adjustment in French adults from who had undergone standardised treatment of complete unilateral cleft lip and palate. Ann Chir Plast Esthet. 2005;50(3):202-5.

43. McWilliams BJ, Paradise LP. Educational, occupational, and marital status of cleft palate adults. Cleft Palate J. 1973; 10:223–229

44. Noar JH. Questionnaire survey of attitudes and concerns of patients with cleft lip and palate and their parents. Cleft Palate Craniofac J. 1991;28:279–284.

45. Sarwer DB, Bartlett SP, Whitaker LA, Paige KT, Pertschuk MJ, Wadden TA. Adult psychological functioning of individuals born with craniofacial anomalies. Plast Reconstr Surg. 1999;103(2):412-8.

46. Madera JM, Hebl MR. Discrimination against facially stigmatized applicants in interviews: an eye-tracking and face-to-face investigation. J Appl Psychol. 2012; 97(2):317–330.

47. Rumsey N, Bull R, Gahagan D. A developmental study of children's stereotyping of facially deformed adults. Br J Psychol. 77. 1986;(77):269–274

48. Hamlet C, Harcourt D. Older adults' experiences of living with cleft lip and palate: a qualitative study exploring aging and appearance. Cleft Palate Craniofac J. 2015; 52(2):e32-40

49. Berger ZE, Dalton LJ. Coping with a cleft II: Factors associated with psychosocial adjustment of adolescents with a cleft lip and palate and their parents. Cleft Palate Craniofac J. 2011;48(1):82-90

50. Feragen KB, Kvalem IL, Rumsey N, Borge AI. Adolescents with and without a facial difference: The role of friendships and social acceptance in perceptions of appearance and emotional resilience. Body Image. 2010;7(4):271-9.

51. Heller A, Tidmarsh W, Pless IB. The psychosocial functioning of young adults born with cleft lip or palate. A follow-up study. Clin Pediatr (Phila). 1981;20(7):459-65.

52. Broder H L, Richman L C. An examination of mental health services for cleft and craniofacial patients. Cleft Palate and Craniofacial J. 1987;24: 158-163

53. Broder H, Strauss RP. Psychosocial problems and referrals among oral-facial team patients. Journal of Oral Rehabilitation. 1991;57(1):31-36.

54. Dutkiewicz-Część cik K. The role of the psychologist as a member of the cleft and craniofacial team in looking after patients and their families from the perspective of the literature and clinical experience. Dev Period Med. 2014;18(1):84-92.

55. Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial anomalies. Cleft Palate Craniofac J. 2017;55:137-156

56. Fisher S, Cleveland SE. Body Image and Personality. New York: Dover; 1968.

57. N. Rumsey, N.M. Stock. Living with a cleft: Psychological challenges, support and intervention. S. Berkowitz (Ed.), Cleft lip and palate. Diagnosis and management, Springer Berlin Heidelberg, Chicago, IL, USA (2013), pp. 907-915

58. Strauss RP, Broder H, Helms RW. Perceptions of appearance and speech by adolescent patients with cleft lip and palate and by their parents. Cleft Palate J. 1988;25(4):335-42.

59. Marcusson A, Paulin G, Ostrup L. Facial appearance in adults who had cleft lip and palate treated in childhood. Scand J Plast Reconstr Surg Hand Surg. 2002;36(1):16-23.

60. Mendelson BK, White DR, Mendelson MJ. Self-esteem and body esteem: effects of gender, age, and weight. J Appl Dev Psychol. 1996;17:321–246.

61. Huang JS, Norman GJ, Zabinski MF, Calfas K, Patrick K. Body image and self-esteem among adolescents undergoing an intervention targeting dietary and physical activity behaviors. J Adolesc Health. 2007;40:245–251.

62. Van Den Elzen ME, Versnel SL, Hovius SE, Passchier J, Duivenvoorden HJ, Mathijssen IM. Adults with congenital or acquired facial disfigurement:

impact of appearance on social functioning. J Craniomaxillofac Surg. 2012;40(8):777-82.

63. Ong J, Clarke A, White P, Johnson M, Withey S, Butler PE. Does severity predict distress? The relationship between subjective and objective measures of appearance and psychological adjustment, during treatment for facial lipoatrophy. Body Image. 2007 ;4(3):239-48.

64. Klassen AF1, Tsangaris E, Forrest CR, Wong KW, Pusic AL, Cano SJ, Syed I, Dua M, Kainth S, Johnson J, Goodacre T. Quality of life of children treated for cleft lip and/or palate: a systematic review. J Plast Reconstr Aesthet Surg. 2012; 65(5):547-57.

65. Kapp K. Self-concept of the cleft lip and/or palate child. Cleft Palate J 1979; 16:171.

66. Berk N W, Cooper M E, Liu Y E, Marazita M L. Social anxiety in Chinese adults with oral-facial clefts. Cleft Palate-Craniofacial J. 2001;38: 126–133.

67. Nicholls W, Harper C, Selvey LA, Robinson S, Hartig G, Persson M. Body Esteem in a Western Australian Cleft Lip and/or Palate Cohort Across 3 Age Groups. Cleft Palate Craniofac J. 2018;55(4):487-498.

68. Maris CL, Endriga MC, Omnell ML, Speltz ML. Psychosocial adjustment in twin pairs with and without hemifacial microsomia. Cleft Palate Craniofac J. 1999; 36(1):43-50.

69. Broder HL, Smith FB, Strauss RP. Developing a behavior rating scale for comparing teachers' ratings of children with and without craniofacial anomalies. Cleft Palate Craniofac J. 2001;38(6):560-5.

70. Becker DB, Petersen JD, Kane AA, Cradock MM, Pilgram TK, Marsh JL. Speech, cognitive, and behavioral outcomes in nonsyndromic craniosynostosis. Plast Reconstr Surg. 2005;116(2):400-7.

71. Kelleher MO, Murray DJ, McGillivary A, Kamel MH, Allcutt D, Earley MJ. Behavioral, developmental, and educational problems in children with nonsyndromic trigonocephaly. J Neurosurg. 2006;105(5 Suppl):382-4.

72. Bannink N, Maliepaard M, Raat H, Joosten KF, Mathijssen IM. Health-related quality of life in children and adolescents with syndromic craniosynostosis. J Plast Reconstr Aesthet Surg. 2010; 63(12):1972-81.

73. Maliepaard M, Mathijssen IM, Oosterlaan J, Okkerse JM. Intellectual, behavioral, and emotional functioning in children with syndromic craniosynostosis. Pediatrics. 2014 ; 133(6):1608-15.

74. Bannink N, Maliepaard M, Raat H, Joosten KF, Mathijssen IM. Obstructive sleep apnea-specific quality of life and behavioral problems in children with syndromic craniosynostosis. J Dev Behav Pediatr. 2011;32(3):233-8.

75. Krueckeberg SM, Kapp-Simon KA. Longitudinal follow-up of social skills in children with and without craniofacial anomalies. Presented at the Annual Meeting of the American Cleft Palate-Craniofacial Association, 1997; New Orleans, LA.

76. Speltz ML, Morton K, Goodell EW, Clarren SK. Psychological functioning of children with craniofacial anomalies and their mothers: follow-up from late infancy to school entry. Cleft Palate J 1993;30:482-489.

77. Cloonan YK, Collett B, Speltz ML, Anderka M, Werler MM. Psychosocial outcomes in children with and without non-syndromic craniosynostosis: findings from two studies. Cleft Palate Craniofac J. 2013; 50(4):406-13.

78. Dufton LM, Speltz ML, Kelly JP, Leroux B, Collett BR, Werler MM. Psychosocial outcomes in children with hemifacial microsomia. J Pediatr Psychol. 2011; 36(7):794-805

79. Topolski TD, Edwards TC, Patrick DL. Quality of life: how do adolescents with facial differences compare with other adolescents? Cleft Palate Craniofac J. 2005;42(1):25-32.

80. Wehby GL, Ohsfeldt RL, Murray JC. Health professionals' assessment of health-related quality of life values for oral clefting by age using a visual analogue scale method. Cleft Palate Craniofac J. 2006b; 43:383–91.

81. Kapp-Simon KA, Simon DJ, Kristovich S. Self-perception, social skills, adjustment, and inhibition in young adolescents with craniofacial anomalies. Cleft Palate Craniofac J 1992;29:352–356.

82. Damiano P, Tyler M, Romitti P, Momany E, Jones M, Canady J, Karnell M, Murray J. Health-related quality of life among preadolescent children with oral clefts. Pediatrics. 2007; 120:e283–90.

83. Marcusson A, Akerlind I, Paulin G. Quality of life in adults with repaired complete cleft lip and palate. Cleft Palate Craniofac J. 2001;38:379–385.

84. Wogelius P, Gjorup H, Haubek D, Lopez R, Poulsen S. Development of Danish version of child oralhealth-related quality of life questionnaires (CPQ8–10 and CPQ11–14). BMC Oral Health. 2009; 22.9(1):11

85. Oosterkamp BC, Dijkstra PU, Remmelink HJ, van Oort RP, Goorhuis-Brouwer SM, Sandham A, DeBont LG. Satisfaction with treatment outcome in bilateral cleft lip and palate patients. Int J Oral Maxillofac Surg. 2007; 36:890–5.

86. Aravena PC, Gonzalez T, Oyarzún T, Coronado C. Oral Health-Related Quality of Life in Children in Chile Treated for Cleft Lip and Palate: A Case-Control Approach. Cleft Palate Craniofac J. 2017;54(2):e15-e20

87. Geels LM, Kieffer JM, Hoogstraten J, Prahl-Andersen B. Oral health-related quality of life of children with craniofacial conditions. Cleft Palate Craniofac J. 2008;45(5):461-7.

88. Knudsen E, Maltese G, Tarnow P, Tovetjärn R, Kölby L. Parental estimation of early psychological development in children operated on for single suture synostosis. J Plast Surg Hand Surg. 2012;46(3-4):152-4.

89. Prescott T, Redfors M, Rustad CF, Eiklid KL, Geirdal AØ, Storhaug K, Jensen JL. Characterization of a Norwegian cherubism cohort; molecular genetic findings, oral manifestations and quality of life. Eur J Med Genet. 2013;56(3):131-7

90. Raposo-Amaral CE, Raposo-Amaral CA, Garcia Neto JJ, Farias DB, Somensi RS. Apert syndrome: quality of life and challenges of a management protocol in Brazil. J Craniofac Surg. 2012;23(4):1104-8.

91. Raposo-Amaral CE, Neto JG, Denadai R, Raposo-Amaral CM, Raposo-Amaral CA. Patient-reported quality of life in highest-functioning Apert and Crouzon syndromes: a comparative study. Plast Reconstr Surg. 2014;133(2):182e-91e.

92. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders: DSM-IV-TR. Washington, DC: American Psychiatric Association, 2000.

93. Endriga MC, Kapp-Simon KA. Psychological issues in craniofacial care: state of the art. Cleft Palate Craniofac J. 1999 ;36(1):3-11.

94. Singh VP, Moss TP. Psychological impact of visible differences in patients with congenital craniofacial anomalies. Prog Orthod. 2015;16:5.

95. Hutchinson K, Wellman MA, Noe DA, Kahn A. The psychosocial effects of cleft lip and palate in non-Anglo populations: a cross-cultural meta-analysis. Cleft Palate Craniofac J. 2011;48(5):497-508.

96. Nicholls W., Persson M., Robinson S., Selvey L. Psychosocial Impacts of Cleft: A Review of the Literature. AASCIT Journal of Psychology. 2016; 1(2): 14-32.

97. Christensen K, Mortensen PB. Facial clefting and psychiatric diseases: a follow-up of the Danish 1936–1987 Facial Cleft cohort. Cleft Palate Craniofac J. 2002; 39(4):392–6.

98. Hunt O, Burden D, Hepper P, Stevenson M, Johnston C. Parent reports of the psychosocial functioning of children with cleft lip and/or palate. Cleft Palate Craniofac J. 2007;44(3):304-11.

99. Murray L, Arteche A, Bingley C, Hentges F, Bishop DV, Dalton L, Goodacre T, Hill J; Cleft Lip and Palate Study team. The effect of cleft lip on socio-emotional functioning in school-aged children. J Child Psychol Psychiatry. 2010;51(1):94-103.

100. Demir T, Karacetin G, Baghaki S, Aydin Y. Psychiatric assessment of children with nonsyndromic cleft lip and palate. Gen Hosp Psychiatry. 2011;33(6):594-603.

101. Ramstad T, Ottem E, Shaw WC. Psychosocial adjustment in Norwegian adults who had undergone standardised treatment of complete cleft lip and palate: self-reported problems and concerns with appearance. Scand J Plast Reconstr Hand Surg. 1995b;29:329– 336

102. Nicholls W, Harper C, Robinson S, Persson M, Selvey L. Adult-Specific Life Outcomes of Cleft Lip and Palate in a Western Australian Cohort. Cleft Palate Craniofac J. 2018. *In Press.*

103. Feragen KB, Stock NM. Psychological adjustment to craniofacial conditions (excluding oral clefts): A review of the literature. Psychol Health. 2017;32(3):253-288.

104. Tovetjärn R, Tarnow P, Maltese G, Fischer S, Sahlin PE, Kölby L. Children with Apert syndrome as adults: a follow-up study of 28 Scandinavian patients. Plast Reconstr Surg. 2012;130(4):572e-576e.

105. Roberts RM. Discrimination among adults with craniofacial conditions. J Craniofac Surg. 2014;25(1):77-81.

106. Roberts RM, Mathias JL. Predictors of mental health in adults with congenital craniofacial conditions attending the Australian craniofacial unit. Cleft Palate Craniofac J. 2013;50(4):414-23.

107. Feragen KB, Stock NM, Sharratt ND, Kvalem IL. Self-perceptions of romantic appeal in adolescents with a cleft lip and/or palate. Body Image. 2016;18:143-52.

108. Christensen K, Juel K, Herskind AM, Murray JC. Long term follow up study of survival associated with cleft lip and palate at birth. BMJ. 2004; 328:1405–1408.

109. Herskind A, Christensen K, Juel K, Fogh-Anderson P. Cleft lip: a risk factor for suicide. Paper presented at: 7th International Congress on Cleft Palate

and Related Craniofacial Anomalies; November 2, 1993; Broadbeach, Queensland, Australia.

110. Kapp-Simon KA, Simon DJ, Kristovich S. Self-perception, social skills, adjustment, and inhibition in young adolescents with craniofacial anomalies. Cleft Palate Craniofac J. 1992; 29(4):352-6.

111. Berger ZE, Dalton LJ. Coping with a cleft: psychosocial adjustment of adolescents with a cleft lip and palate and their parents. Cleft Palate Craniofac J. 2009;46(4):435-43

112. Patel Z, Ross E. Reflections on the cleft experience by South african adults: use of qualitative methodology. Cleft Palate Craniofac J. 2003;40(5):471-80.

113. Drotar D, Baskiewicz A, Irvin N, Kennell J, Klaus M. The adaptation of parents to the birth of an infant with a congenital malformation: a hypothetical model. Pediatrics. 1975 Nov;56(5):710-7.

114. Nelson P, Glenny AM, Kirk S, Caress AL. Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. Child Care Hlth Dev. 2012;38(1):6–20.

115. Nelson J, O'Leary C, Weinman J. Causal attributions in parents of babies with a cleft lip and/or palate and their association with psychological well-being. Cleft Palate Craniofac J. 2009;46(4):425-34.

116. Johansson B, Ringsberg KC. Parents' experiences of having a child with cleft lip and palate. J Adv Nurs. 2004 Jul;47(2):165-73.

117. Bradbury ET, Hewison J. Early parental adjustment to visible congenital disfigurement. Child Care Health Dev. 1994 Jul-Aug;20(4):251-66.

118. Kramer FJ, Baethge C, Sinikovic B, Schliephake H. An analysis of quality of life in 130 families having small children with cleft lip/palate using the impact on family scale. Int J Oral Maxillofac Surg. 2007;36(12):1146-52.

119. Pope AW, Tillman K, Snyder HT. Parenting stress in infancy and psychosocial adjustment in toddlerhood: a longitudinal study of children with craniofacial anomalies. Cleft Palate Craniofac J. 2005;42(5):556-9.

120. Thamilselvan P, Kumar M S, Murthy J, Sharma MK, Kumar N R. Psychosocial issues of parents of children with cleft lip and palate in relation to their behavioral problems. J Cleft Lip Palate Craniofac Anomal 2015;2:53-7

121. Crerand CE, Kapa HM, Litteral J, Pearson GD, Eastman K, Kirschner RE. Identifying Psychosocial Risk Factors Among Families of Children With

Craniofacial Conditions: Validation of the Psychosocial Assessment Tool-Craniofacial Version. Cleft Palate Craniofac J. 2018;55(4):536-545.

122. Rumsey N, Harcourt D. Visible difference amongst children and adolescents: issues and interventions. Dev Neurorehabil. 2007;10(2):113–123.

123. Baker SR, Owens J, Stern M, Willmot D. Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. Cleft Palate Craniofac J. 2009;46(3):229-36

124. Kapp-Simon KA, McGuire DE, Long BC, Simon DJ. Addressing quality of life issues in adolescents: social skills interventions. Cleft Palate Craniofac J. 2005;42(1):45-50.

125. Strauss RP. “Only skin deep”: health, resilience, and craniofacial care. Cleft Palate Craniofac J. 2001; 38:226–230.

126. Stock NM, Feragen KB, Rumsey N. "It Doesn't All Just Stop at 18": Psychological Adjustment and Support Needs of Adults Born With Cleft Lip and/or Palate. Cleft Palate Craniofac J. 2015;52(5):543-54.

127. Loewenstein J, Sutton E, Guidotti R, Shapiro K, Ball K, McLean D, Biesecker B. The art of coping with a craniofacial difference: helping others through "Positive Exposure". Am J Med Genet A. 2008;146A(12):1547-57

128. Strauss RP, Fenson C. Experiencing the "good life": literary views of craniofacial conditions and quality of life. Cleft Palate Craniofac J. 2005;42(1):14-8.

129. Thompson RA and Broom L (2009) Positively managing intrusive reactions to disfigurement: an interpretative phenomenological analysis of naturalistic coping. Diversity in Health and Care, 6: 171–80.

130. Eiserman W. Unique outcomes and positive contributions associated with facial difference: expanding research and practice. Cleft Palate Craniofac J. 2001;38(3):236-44.

131. Stock NM, Feragen KB, Rumsey N. Adults' Narratives of Growing up With a Cleft Lip and/or Palate: Factors Associated With Psychological Adjustment. Cleft Palate Craniofac J. 2016;53(2):222-39.

132. Klein T, Pope AW, Getahun E, Thompson J. Mothers' reflections on raising a child with a craniofacial anomaly. Cleft Palate Craniofac J. 2006 Sep;43(5):590-7.

133. Kelton RW. Facing up to stigma: workplace and personal strategies. Cleft Palate Craniofac J. 2001;38(3):245-7.

134. Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: a systematic review. Eur J Orthod. 2005; 27(3):274–85

135. Wehby GL, Cassell CH. The impact of orofacial clefts on quality of life and healthcare use and costs. Oral Dis. 2010;16:3–10

136. Kapp-Simon K. Psychological issues in cleft lip and palate. Clin Plast Surg. 2004;31:347–352

137. Bjerke SM, Feragen KB, Bergvik S. Strengths and Difficulties Questionnaire (SDQ): Informant Agreement Between Children Born With Cleft Lip and/or Palate and Their Parents. Cleft Palate Craniofac J. 2018;55(2):204-212.

138. Feragen KB, Stock NM. When there is more than a cleft: psychological adjustment when a cleft is associated with an additional condition. Cleft Palate Craniofac J. 2014;51(1):5-14.

139. Van Roy B, Grøholt B, Heyerdahl S, Clench-Aas J. Understanding discrepancies in parent-child reporting of emotional and behavioral problems: effects of relational and socio-demographic factors. BMC Psychiatry. 2010;10:56-68.

140. Achenbach TM, McConaughy SH, Howell CT. Child/adolescent behavioral and emotional problems: implications of crossinformant correlations for situational specificity. Psychol Bull. 1987;101:213-232.

141. Goodman R (1997) The Strengths and Difficulties Questionnaire: A Research Note. Journal of Child Psychology and Psychiatry, 38, 581-586.

142. Goodman, R. The extended version of the Strengths and Difficulties Questionnaire as a guide to child psychiatric caseness and consequent burden. Journal of Child Psychology and Psychiatry. 1999;40(5),791 – 799.

143. Van Roy B, Grøholt B, Heyerdahl S, Clench-Aas J. Understanding discrepancies in parent-child reporting of emotional and behavioral problems: effects of relational and socio-demographic factors. BMC Psychiatry. 2010;10:56-68.

144. Offord DR, Boyle MD, Racine Y, Szatmari P, Fleming JE, Sanford M, Lipman EL. Integrating assessment data from multiple informants. J Am Acad Child Adolesc Psychiatry. 1996;35:1078-1085.

145. Achenbach TM, McConaughy SH, Howell CT. Child/adolescent behavioral and emotional problems: implications of crossinformant correlations for situational specificity. Psychol Bull. 1987;101:213-232.

146. Goodman R, Scott S. Comparing the Strengths and Difficulties Questionnaire and the Child Behavior Checklist: is small beautiful? J Abnorm Child Psychol. 1999;27(1):17-24.

147. Goodman R. Psychometric properties of the strengths and difficulties questionnaire. J Am Acad Child Adolesc Psychiatry. 2001;40(11):1337-45.

148. Goodman R, Ford T, Simmons H, Gatward R, Meltzer H. Using the Strengths and Difficulties Questionnaire (SDQ) to screen for child psychiatric disorders in a community sample. Br J Psychiatry. 2000;177:534-9.

149. Goodman R, Renfrew D, Mullick M. Predicting type of psychiatric disorder from Strengths and Difficulties Questionnaire (SDQ) scores in child mental health clinics in London and Dhaka. Eur Child Adolesc Psychiatry. 2000;9(2):129-34.

150. Warnick, E. M., M. B. Bracken and S. Kasl. "Screening efficiency of the Child Behavior Checklist and Strengths and Difficulties Questionnaire: A systematic review." Child and Adolescent Mental Health, 2008;13(3): 140-147

151. Vostanis P. Strengths and Difficulties Questionnaire: research and clinical applications. Curr Opin Psychiatry. 2006;19:367-372.

152. Feragen KB, Stock NM, Rumsey N. Toward a reconsideration of inclusion and exclusion criteria in cleft lip and palate: implications for psychological research. Cleft Palate Craniofac J. 2014;51(5):569-78

THESIS STATEMENT AND PURPOSE

The primary purpose of the current study is to identify and compare the difficulties and strengths among 3 groups of adolescents: adolescents with Craniofacial conditions other than cleft lip and/or palate (CFA), adolescent with cleft lip and/or palate (CLP), and controls (orthodontic patients without CFA or Cleft lip and palate). Our secondary objectives are to identify social factors that may influence their adjustments levels, and to compare the informant agreement among parent, teacher and self-reports.

The main objective of this study is attempt to address some of current gaps in the vast body of literature concerning the psychosocial adjustments of individuals with CFA. As mentioned in our literature review, previous studies tended to focus more on patients with CLP, while the data on other CFA is still scarce. The lack of appropriate control groups has been highlighted, along with an inconsistency of the instruments used, which made it challenging to draw definite conclusions regarding the affected individuals. Moreover, many researchers have voiced the need for more studies that focus on coping mechanisms and strengths of this population, rather than just the deficits. Finally, while teacher observations have been widely utilized by social scientists to measure children's psychological adjustment, their presence in the craniofacial literature is scarce, which could be attributed to the practical limitations of gathering these reports.

In an effort to overcome the aforementioned limitations of previous studies, we decided to include 3 groups of participants, CFA (other than CLP), CLP, and controls. Moreover, the primary instrument employed in this study is the extended version of the Strengths and Difficulties Questionnaire (SDQ), with the impact supplement. The SDQ was chosen because it is a brief, yet vastly utilized instrument, with satisfactory reliability and validity. Furthermore, the SDQ includes 4 subscales for difficulties, and 1 for strengths, allowing us to look into specific subcategories of the psychosocial adjustment, rather than a generalized number. We included the Impact Supplement of the SDQ to help us understand the perceived impact of the symptoms that may be exhibited by participants, and to add to the body of research that has previously utilized the SDQ without the impact supplement. For each subject, we attempted to collect the questionnaires from 3 informants, (self-reports, a parent, and a teacher). Former investigators have highlighted the importance of using multi-informant studies, in order to obtain a comprehensive, diverse set of data that would allow us to draw more accurate conclusions. While numerous studies have gathered data from patients and parents, teacher reports in the craniofacial research remain to be scarce. In contrast to most parents, teachers offer an adult's view of the child in relation to a relatively large peer group of the same age, and social encounters outside home can be more difficult for the affected patients, and thus supports the notion that teacher's reports are valuable. Finally, we added specific questions to the parent questionnaire, in an attempt to explore the

impact of some social support structures on adjustment levels. The structures we explored were, 1) Number of patients living in the household where the subject lives, 2) Number of siblings, and 3) Whether the subject had at least one good friend.

Hypotheses:

Hypothesis #1: There is a significant difference between the Strengths and Difficulties Questionnaire (SDQ) scores among adolescents with craniofacial conditions other than cleft lip and/or palate (CFA), adolescent with cleft lip and/or palate (CLP), and a control group of adolescents seeking orthodontic treatment.

Hypothesis #2: High levels of agreement are found among the SDQ scores as reported by self-evaluations, parents, and teachers.

Hypothesis #3: Social factors, such as number of parents living in the same household, number of siblings, having a good friend, and bullying can significantly influence the total difficulties score.

In conclusion, our proposed study aims at bridging some of the current gaps in the literature, by including a craniofacial anomalies group besides cleft lip and palate, including appropriate controls, including teacher reports along with a multi-informant approach, and using a standardized questionnaire (The strengths

and difficulties questionnaire - SDQ) with established high reliability, validity, sensitivity, and specificity.

Our findings would allow us to offer a better understanding of the complex psychosocial interactions of the affected individuals, and the influence of some social factors on adjustment levels, with the long-range goal of developing evidence-based intervention strategies by school personnel and pediatric psychologists to provide this vulnerable population with their specific, optimal needs.

MANUSCRIPT FOR PUBLICATION

ABSTRACT

Background: Numerous studies have investigated psychosocial adjustment levels among adolescents with cleft lip and/or palate, however, our understanding of other craniofacial conditions remains to be scarce. The primary purpose of the current study is to bridge some of the gaps in the literature through comparing psychosocial adjustments among three groups of adolescents: craniofacial conditions other than cleft lip and/or palate (CFA), adolescent with cleft lip and/or palate (CLP), and a control group (CON). Secondary objectives are identifying social factors that may influence adjustments levels, and comparing informant agreements among parent, teacher and self-reports.

Design: Cross-sectional, multi-informant, controlled survey study.

Participants: 11-17 year olds. 49 with CFA other than CLP, 42 with isolated CL/P, and 55 controls. Total = 146.

Outcome measures: The Strengths and Difficulties Questionnaire (self-report, parent report, and teacher report).

Results: Parent reports showed significant differences among the 3 groups, with CFA exhibiting significantly higher total difficulty scores than CLP (p=0.027) or CON

(p=<0.001), and CLP significantly higher than controls (p=0.016). Subscales showed that CFA had higher peer problems compared to CLP (p=0.001), and CLP was higher than CON (p=0.05). CFA group displayed a significantly higher frequency of abnormal psychosocial adjustment (34.7%), compared to 17% among CLP group, and only 3.8% for controls.

Informant agreements were moderate, yet significant, for parent reports vs self-reports, and parent reports vs teacher reports. However, correlations between self-reports vs teacher reports were not significant. The most predictive factor was the frequency of being bullied, followed by having a good friend. However, the number of parents in the household or the number of siblings were not significant. The four studied social factors combined explained 44.2% of the variance.

Conclusions: A general trend was observed where CFA group displayed the highest psychosocial difficulties, followed by CLP, and the lowest were the CON group. Within the subscales, peer problems constituted the biggest challenge that faces both CFA and CLP groups. Social factors, especially being bullied or having a good friend, could significantly mediate and influence the psychosocial adjustment levels.

KEY WORDS: cleft lip and palate, craniofacial conditions, psychosocial adjustment, informant agreement, SDQ.

INTRODUCTION

Centuries ago, humans did not possess our current scientific knowledge and thus, giving birth to a child with congenital deformities was interpreted by myth, religion and superstition.[1] Infants with congenital deformities were presumed to be a punishment from the gods, or to harbor evil spirits.[2] Such beliefs have led many ancient cultures to the tragic acts of abandonment, exposure, or killing of the affected newborns.[3] Shockingly, even some of the greatest philosophers of all time, such as Plato and Aristotle, advocated these practices, arguing that it will benefit the society by eliminating evil spirits.[4,5] In his *Politics*, Aristotle wrote "*let there be a law that no deformed child shall live.*"[4]

With the advent of modern medicine, these superstitious beliefs and rituals have diminished to a large extent, especially in Western cultures. The current advancements in medicine have tremendously progressed to explain the complex embryological nature of these deformities and to provide the patients with sophisticated healthcare measures in efforts to not just restore the functional aspects of their conditions, but to also normalize their appearance in hopes of improving their quality of life and psycho-social experiences. Today, the term "craniofacial anomalies" (CFA) refers to a diverse group of congenital deformities that affect the

head and the facial structures.[6,7] They are described as "anomalies", because they deviate from what is considered to be normal. In spite of the substantial advancements in the management of patients with CFA, the societal attitudes towards people with craniofacial anomalies have remained largely static, burdening them with the stigma that accompanies their deformities throughout their lives, as our basic human instincts remain to tarnish our societal attitudes and acceptance of the affected individuals.[8-16]

Erving Goffman, one of the most influential modern sociologists, defined stigma as "*The phenomenon whereby an individual with an attribute which is deeply discredited by his/her society is rejected as a result of the attribute. Stigma is a process by which the reaction of others spoils normal identity.*"[17] The association between appearance and social stereotyping is established in social science research.[18] The implications of the social stigmas that accompany these conditions may reflect adversely on the psycho-social well-being and quality of life of the affected individuals.[19,20,21] This is complicated further by the negative self-perception of the affected individuals, leading to feelings of dissatisfaction, worthlessness and social isolation.[20,21,22] A number of studies found that children with CFA displayed greater social inhibitions, social marginalization, lower self-concept, and were more sad or angry than their peers.[23-27] Tragically, the prevalence of teasing among children with CFA was found to be significantly higher than the ranges for the general population in the United States.[28-35] The implications may extend into adulthood, as

adults with CFA reported being discriminated against, decreased social support, less engagement in social activities, and lower likelihood of being married or employed.[36-42]

Negative social experiences and a poor body image were concluded to be major psychological risk factor for adolescents with craniofacial conditions.[43,44,45,46,47] A number of studies have shown that subjects with CFA or CLP tend to have a lower self-concept, lower quality of life, higher levels of behavioral problems, and an increased risk of psychosocial maladjustments when compared to non-affected individuals.[20,21,28,39,48-67] Other studies found that affected individuals may have an elevated risk of developing psychopathologies, social anxiety, depression and substance abuse.[36,59,68-73] Two particularly alarming studies reported an increase in mortality and a doubling of suicide rates among adults with CLP.[74,75] However, these findings were inconsistent in the literature, as other studies found normal or higher levels of psycho-social coping in CFA population compared to non-affected peers, , which may be attributed to increased emotional resilience.[44,76-89] In a classic systematic review, Endriga and Kapp-Simon deduced that although the research suggests that many children with CFA do not experience psychological problems, it is evident that they encounter a higher risk.[66] In the same vein, a recent meta-analysis concluded that irrespective of age, gender, or culture, individuals with CLP have lower psychosocial functioning than non-affected individuals.[67] This was supported by another recent systematic review by Nicholls et al, where 89% of the papers they

reviewed pointed towards an elevated risk of developing psychosocial difficulties for children with clefts.[68]

More recently, investigators have shifted their focus towards exploring the strengths of this population and factors that may influence their adjustment levels. Studies reported that, among many beneficial coping skills, social competence and social support were major predictors of adjustment levels.[43,44,63,66,89-97] A remarkable study by Eiserman revealed that half of his sample would not choose to remove the experience of having a facial difference from their lives or their children's lives, if given the opportunity.[89]

Systematic reviews as recent as 2017 have attributed the inconsistency among the findings in this field of research to the lack of appropriate control groups, scarcity of studies that focus on CFA other than CLP, the inconsistency among instruments used, variability of informants, diversity of CFA, the lack of longitudinal studies, and the lack of studies that explore why some affected individuals may cope better than others. [98,99,100]

Due to the wide variety of instruments that have been utilized in this field of research, researchers have pointed out the need for more uniform studies that utilize similar instruments. The Strengths and Difficulties Questionnaire (SDQ) was developed by Dr. Robert Goodman in 1997 as a brief screening tool for psychosocial assessment of children and adolescents.[101] Since then, the SDQ has become one of the most extensively employed instruments in psychological assessment, exhibiting

various version for multiple informants, satisfactory validity and reliability, as well as a high specificity and moderate sensitivity in predicting child psychiatric diagnosis, making it a valuable screening instrument.[101-108] In craniofacial research, the SDQ has recently been utilized by a number of investigators, and since 2009, has been adopted by the Oslo craniofacial team as their instrument of choice for routine psychological assessment.[43,86,109-111]

The primary purpose of the current study is to gain a better understanding of the psychosocial adjustments of individuals with CFA or CLP, through bridging some of the aforementioned gaps in the literature. We aim to utilize the SDQ to compare the psychosocial adjustment levels among three groups of adolescents:

1- Craniofacial conditions other than cleft lip and/or palate (CFA), 2- Isolated cleft lip and/or palate (CLP), and 3- Non-Affected Controls, in an attempt to answer the following questions:

1- Are adolescents with CFA at a higher risk of having psychosocial difficulties compared to individuals with CLP or controls?
2- Is there an agreement among SDQ scores as reported by self-evaluations, parents, and teachers?
3- Which social factors have the largest influence on psychosocial adjustment levels?

MATERIALS AND METHODS

The design of this cross-sectional, multi-informant, controlled survey study was approved by the sponsoring institution's Institutional Review Board (UHCMC IRB number: 07-17-39). Three groups of participants (Total n=146) were recruited into this study as follows; 1- CFA group (adolescents with craniofacial conditions excluding isolated cleft lip and/or palate) (n=49), 2- CLP group (adolescents with cleft lip and/or palate) (n=42), and 3- CON group (control group of adolescents seeking orthodontic treatment) (n=55). Subjects with CL/P plus other craniofacial conditions were grouped under CFA. The first 2 groups (CFA and CLP) were consecutively recruited from 2 institutions; the craniofacial orthodontic clinic at Case Western Reserve University and the University Hospitals Cleveland Medical Center craniofacial team, while the CON group were recruited from the Case Western Reserve University orthodontic core clinic.

Inclusion criteria: 1. CFA group: Ages 11-17. With syndromic or non-syndromic craniofacial conditions (excluding isolated cleft lip and/or palate), 2. CLP group: Ages 11-17. Cleft lip and/or palate without any other craniofacial conditions, 3. CON group: Ages 11-17. Orthodontic patients not affected with CFA or CLP.

Exclusion criteria: 1- Subjects that were considered by their parents to be developmentally unfit to complete the questionnaire, 2- Parents or teenagers who declined to participate.

We attempted to collect three versions of the SDQ for each subject, one self-report, one from a parent and one from a teacher (recommended by the subject or parents). Two additional questions were added to the parent questionnaire to analyze support structures: 1- Number of parents in the household, 2- Number of siblings. All the study data were collected electronically using tablets or through e-mail, and were stored securely on REDCap electronic data capture tools hosted at University Hospitals Cleveland Medical Center.[112] Recruitment was conducted as follows: parents and patients were approached during their routine visits to the craniofacial team/orthodontic clinic. After agreeing to participate and verbally consenting, they were given a tablet with a link to the UH REDCap web portal for their respective survey. Moreover, a report was run on our database to identify CFA and CLP subjects that match our inclusion criteria, and an e-mail was sent out to the eligible individuals with the study code, and a link to the REDCap portal to fill the survey. The parent survey had a question about whether they agree to include a teacher, and if they accept, they were prompted to enter the name of a teacher and their respective e-mail address. Declining teacher participation did not exclude them from the study. Subsequently, e-mails were sent to teachers with the link to the survey, along with the specific subject code. If the parents failed to include a teacher e-mail, a sealed envelope addressed to the teacher was sent to the school, with instructions and a barcode that would link them to the study. Informed consents and child assent forms were available as attachments on the respective online surveys. To

encourage participation, each participating teenager was sent a digital $10 gift card as a compensation.

Statistical analysis was conducted using IBM SPSS version 25 (IBM, Armonk, NY) to compare the overall SDQ score, and each of the five subscales individually. The SDQ consists of 25 items grouped into 5 subscales: emotional symptoms, conduct problems, hyperactivity, peer relationships, and prosocial behavior.[101] Each item is scored ‘‘not true,’’ ‘‘somewhat true,’’ or ‘‘certainly true’’ (score range, 0 to 2).[101] The first four subscales are added together to generate a total difficulties score (20 items with a score range of 0 to 40), while the last subscale addresses the strengths (five items with a score range of 0-10). For the difficulties, a higher score indicates increased risk for psychological problems, while for the strengths, a higher score indicates greater adjustment levels. In addition, the impact supplement is composed of five additional questions that identify the overall psychological impact on different aspects of the adolescent’s life, with a score that ranges from 0 to 10 for parent- and self-report, and from 0 to 6 for teacher-report.[102] Moreover, the SDQ scores were categorized into three bands of psychosocial adjustment levels, ‘normal’, ‘borderline’ and ‘abnormal’. In the present study, we collected parent, teacher and self-reports to compare the total SDQ scores, each of the subscales individually, the impact supplement, and the categorical grouping among our three groups of subjects (CFA, CLP, and CON). In addition, we compared the agreement among the SDQ scores of the three informant groups (self, parents, teachers). Finally, we

analyzed the influence of four social factors on the total difficulty score, as reported by parents: 1-Number of parents living in the same household, 2-Number of siblings, 3-Having one good friend, and 4-Frequency of being bullied or teased.

Statistical Design

The computation of the sample size was based on a power analysis, using the total difficulty score means from a similar study. A sample size test was run and concluded that with a sample of at least 42 subjects per group, our study would yield a power of 95%, with an α-level of <0.05. SPSS statistical analysis software, version 25 (IBM, Armonk, NY) was used for statistical analysis. First, we ran an independent T-test to identify any gender differences for the total parent SDQ scores, since no significant differences were found, we combined males and females into one group. Second, a Shapiro-Wilk test was used to determine the normality of distribution of the total SDQ scores. Since a normal distribution was observed (p= >0.05), we utilized parametric tests for our study as follows:

1- One-way repeated measures ANOVA to compare total score, each subscale, and the impact supplement among groups, and a Tukey Post-Hoc test was used to identify where the differences are, if any. 2- The categorical comparisons among the groups were done using Chi-square test to identify and compare the percentage of normal, borderline, or abnormal SDQ scores among each group. 3- Informant agreements were analyzed using Pearson correlation coefficient, and

multiple paired T-tests, to compare parent, teacher and self-reports. 4- Finally, we ran a Multiple Regression analysis to determine the influence of four social factors (number of siblings, number of parents in the household, having at least one good friend, and frequency of being bullied) on the total difficulties score, as reported by parents.

With this in mind, we aim to answer the following questions: 1- Are there any significant differences in the total SDQ score, the subscales, or the impact supplement among the 3 groups?, 2-What are the agreement levels among parent, teacher and self-reports?, and 3- Which of the four studied social factors have the biggest influence on their psychosocial functioning, based on parent reports?

RESULTS

Descriptive statistics of our study population are presented in Table 1. Males (43.2%) and females (56.8%) were combined into one group for statistical analysis since no significant differences were found between genders for any of the 3 groups. The total sample included 49 CFA (mean age=12.5), 42 CLP (mean age=12.3) and 55 CON (mean age=13.8), (Total = 146) (Mean age=12.9). We gathered 144 self-reports, 142 parent reports, and 86 teacher reports. The slight mismatch among the number of parent and self-reports was due to some incomplete questionnaires, while the attrition of the teachers' sample was attributed to parents unwilling to include

teachers (n=15), lack of response from teachers (n=29) or homeschooled participants (n=12).

Table 1: Demographics

		CFA	CLP	CON	Total	Sig.†
Number of subjects	Parent reports	49	41	52	142	
	Self-reports	49	40	55	144	
	Teacher reports	26	23	37	86	
	Total	49	42	55	146	
Gender	Male	44.9%	45%	47.3%	43.2%	**NS**
	Female	55.1%	55%	52.7%	56.8%	
Mean age		125	12.3	13.8	12.9	**NS**

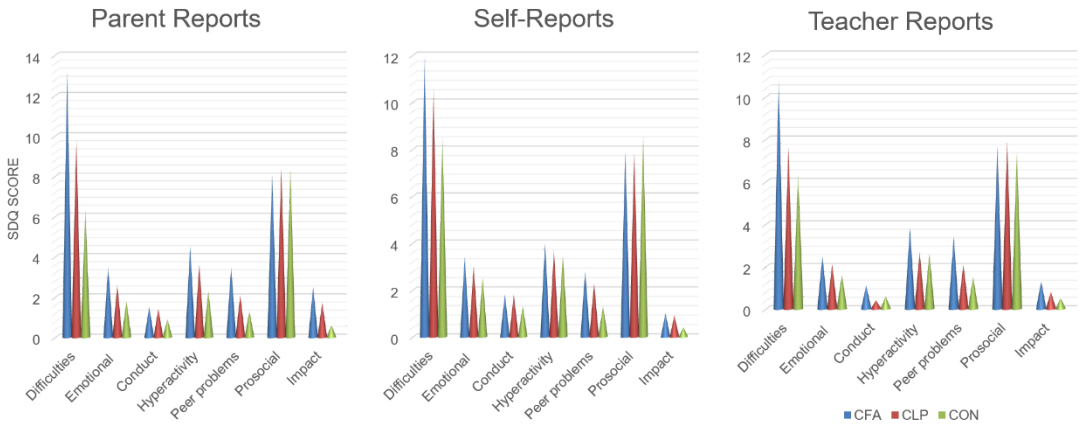
† = One way ANOVA to identify significant differences in the distribution of age or gender among the groups
NS= Not statistically significant

The SDQ score comparisons among the 3 groups for parent reports, self-reports and teacher reports are displayed in Tables 2, 3 and 4, respectively. Parent reports showed significant differences for the total difficulty scores among the 3 groups, with CFA exhibiting significantly higher total difficulty scores than CLP ($p=0.027$) or CON ($p=<0.001$) groups, while CLP were significantly higher than controls ($p=0.016$) (Table 3). Examining the subscales, parents of adolescents with CFA reported significantly higher scores compared to CON for emotional problems

(p=0.001), hyperactivity (p=<0.001), and peer problems (p=<0.001). Moreover, CFA had higher scores of peer problems compared to CLP (p=0.001), and CLP was higher than CON (p=0.04). No significant differences were found for conduct problems or prosocial behavior among any of the groups. The impact supplement revealed a significantly higher psychosocial impact on the life of adolescents with CFA compared to CON (p=<0.001), (Table 2). A general trend was observed where the mean difficulty scores across all the subscales, except prosocial behavior, showed a steady increase going from CON to CFA, with CLP being in the middle (Fig. 1).

Significant findings from the self-reports were slightly different, although the means represented a similar pattern (Fig. 1). The only significant differences for the total difficulty score were between CFA and CON (p=0.004), while the only significantly different subscale was peer problems, with CFA higher than CON (p=<0.001), and CLP higher than CON (p=0.011). Self-reports showed no significant differences for the impact supplement (Table 3).

Figure 1. Mean SDQ scores for all groups



A similar trend is observed across all three informant groups, for all subscales (except prosocial), where the highest levels of difficulties can be observed for the CFA group, followed by CLP, while CON group has the lowest scores.

Table 2: Parent reports (n=142) - SDQ scores and subscales

	CFA (n=49)	CLP (n=41)	CON (n=52)	Sig. †	Tukey Post-Hoc		
	Mean± SD	Mean ± SD	Mean±SD		Group	Mean difference	Sig.*
Total Difficulties	13.1 ± 6.7	9.8 ± 6.3	6.3 ± 4.6	<0.001**	CFA vs CLP	3.3	0.027*
					CFA vs CON	6.8	<0.001**
					CLP vs CON	3.5	0.016*
Emotional problems	3.5 ± 2.4	2.6 ± 2.3	1.8 ± 2.0	<0.001**	CFA vs CON	1.7	<0.001**
Conduct problems	1.5 ± 1.8	1.4 ± 1.7	0.9 ± 1.2	.115			
Hyperactivity	4.5 ± 2.9	3.6 ± 2.7	2.3 ± 2.4	<0.001**	CFA vs CON	2.2	<0.001**
Peer Problems	3.5 ± 1.8	2.1 ± 2.1	1.3 ± 1.1	<0.001**	CFA vs CLP	1.4	<0.001**
					CFA vs CON	2.2	<0.001**
					CLP vs CON	0.8	0.04*
Prosocial	8.1 ± 1.8	8.4 ± 1.7	8.3 ± 1.7	0.69			
Impact on QOL	2.5 ± 2.8	1.7 ± 2.1	0.6 ± 1.5	<0.001**	CFA vs CON	1.9	<0.001**

N= number of subjects
† = One way ANOVA to compare the means among the groups
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.001

Table 3: Self reports (n=144) - SDQ scores and subscales

		CFA (n=49)	CLP (n=40)	CON (n=55)	Sig. †	Tukey Post-Hoc for significant findings		
		Mean ± SD	Mean ± SD	Mean±SD		Group	Mean difference	Sig.*
Total Difficulties		12.0 ± 6.2	10.6 ± 5.4	8.5 ± 1.7	0.005*	CFA vs CON	3.5	0.004*
Emotional problems		3.4 ± 2.6	3 ± 2.2	2.5 ± 2.1	.101			
Conduct problems		1.8 ± 1.7	1.8 ± 1.5	1.3 ± 1.5	0.142			
Hyperactivity		4.0 ± 2.3	3.7 ± 1.9	3.5 ± 2.2	0.459			
Peer Problems		2.8 ± 1.8	2.3 ± 1.7	1.3 ± 1.2	<0.001**	CFA vs CON	1.4	<0.001**
						CLP vs CON	0.9	0.011*
Prosocial		7.9 ± 2.1	7.8 ± 1.8	8.5 ± 1.7	0.145			
Impact on QOL		1.0 ± 1.7	0.9 ± 1.7	0.4 ± 0.9	0.091			
	N= number of subjects † = One way ANOVA to compare the means among the groups *= Statistically significant at a p-value of <0.05 **= Statistically significant at a p-value of <0.001							

Meanwhile, teacher reports displayed similar findings to self-reports, where the only significant differences were between CFA and CON for the total difficulty score (p=0.026), and peer problems (p=0.003). (Table 4).

Table 4. Teacher reports (n=86) - SDQ scores and subscales

	CFA (n=26)	CLP (n=23)	CON (n=37)	Sig.†	Tukey Post-Hoc for significant findings		
	Mean± SD	Mean± SD	Mean±SD		Groups	Mean difference	Sig.*
Total Difficulties	10.8 ±7.9	7.6 ± 5.7	6.3 ± 5.8	0.030 *	CFA vs CON	4.4	0.026 *
Emotional problems	2.5 ± 2.5	2.1 ± 2.0	1.6 ± 2.0	.261			
Conduct problems	1.1 ± 1.7	0.4 ± 0.8	0.6 ± 1.2	0.136			
Hyperactivity	3.8 ± 2.9	2.7 ± 2.7	2.6 ± 3.1	0.288			
Peer Problems	3.4 ± 2.6	2.1 ± 2.4	1.5 ± 1.7	0.004 **	CFA vs CON	1.9	0.003 **
Prosocial	7.7 ± 2.3	7.9 ± 2.5	7.4 ± 2.1	0.680			
Impact on QOL	1.3 ± 1.7	0.8 ± 1.5	0.5 ± 1.1	0.098			

N= number of subjects
† = One way ANOVA to determine significant differences among the groups
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.01

Although SDQ scores can be used as continuous variables, it is sometimes helpful to categorize scores. The scores for each individual were grouped as "Normal", "Borderline", and "Abnormal", according to baseline data reported by Robert Goodman.[101]. Parent reports showed that only 59.2% of CFA exhibited a

normal behavior, compared to 73.2% of CLP, and 94.2% of CON. Borderline behavior was 6.1% for CFA, 9.8% for CLP, and 1.9% for CON. One alarming finding was that 34.7% of CFA exhibited abnormal levels of adjustment, in comparison to 17.1% of CLP and only 3.8% of CON showed abnormal results (Table 5) (Fig. 2). Chi-square test revealed that these differences were statistically significant (p=0.001). A similar statistically significant pattern was found for self-reports (p=0.015). Surprisingly, teacher reports showed no statistically significant differences for the categorical distributions across the 3 groups, although the trends were similar (Table 6).

Hence, it may be safe to conclude that, within our sample, CFA displayed the highest levels of psychosocial problems, while CON was the lowest. Within the subscales, peer problems seemed to be the biggest challenge that faces both CFA and CLP groups. However, prosocial behavior was almost identical across the 3 groups.

Figure 2. Categorical distributions among the groups

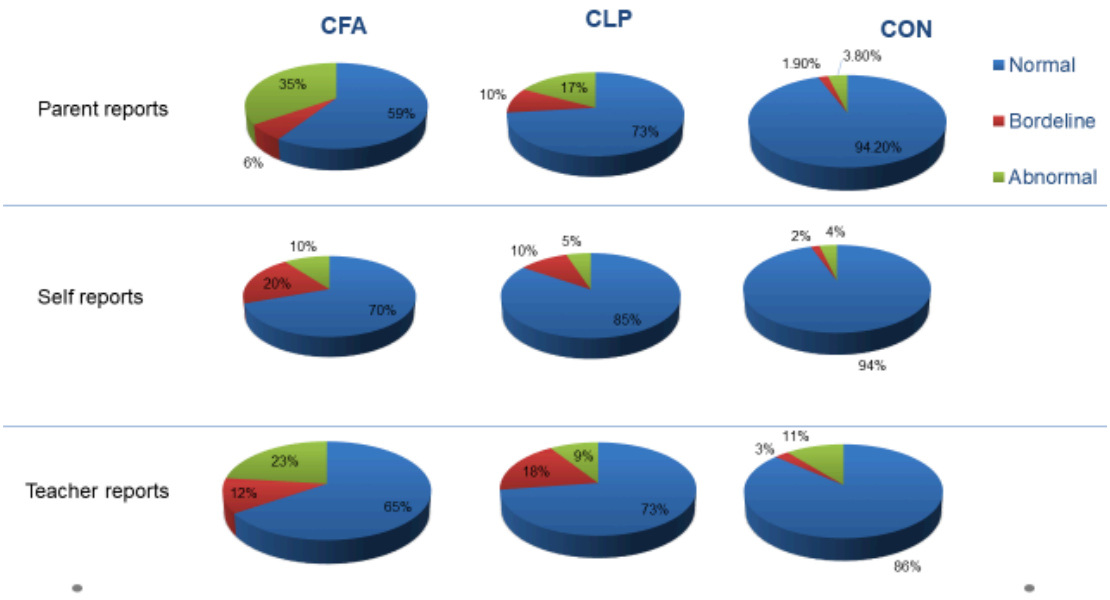


Table 5. Frequencies of normal, borderline, and abnormal SDQ scores

Informant	Category		Group			Total	Sig†
			CFA	CLP	CON		
Parent Reports	Normal	Count	29	30	49	108	**<0.001****
		% in Group	59.2%	73.2%	94.2%	76.1%	
	Borderline	Count	3	4	1	8	
		% in Group	6.1%	9.8%	1.9%	5.6%	
	Abnormal	Count	17	7	2	26	
		% in Group	34.7%	17.1%	3.8%	18.3%	
Self Reports	Normal	Count	34	33	52	119	**0.015***
		% in Group	69.4%	84.6%	94.5%	83.2%	
	Borderline	Count	10	4	1	15	
		% in Group	20.4%	10.3%	1.8%	10.5%	
	Abnormal	Count	5	2	2	9	
		% in Group	10.2%	5.1%	3.6%	6.3%	
Teacher Reports	Normal	Count	17	16	32	65	**0.147**
		% in Group	65.4%	72.7%	86.5%	76.5%	
	Borderline	Count	3	4	1	8	
		% in Group	11.5%	18.2%	2.7%	9.4%	
	Abnormal	Count	6	2	4	12	
		% in Group	23.1%	9.1%	10.8%	14.1%	

† = Chi-Square test for categorical comparisons
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.001

Our second objective was to determine the level of agreement across the 3 informant groups (Table 6). For the total difficulty score, parent and teens showed the highest levels of agreement with statistically significant, yet moderate, correlations (Pearson correlation=0.59, p=<0.01). The results of the paired T-test (n=140) confirmed these findings, as no significant differences were noted (p=0.164). The second pair, parents vs teachers (n=86), showed similar findings, although the correlation was slightly lower, (Pearson correlation =0.48, p=<0.01), and the paired T-test again showed no significant differences (p=0.117). Interestingly, the third pair, teens vs teachers (n=86), displayed poor levels of agreement, and did not significantly correlate to each other (Pearson correlation=0.13). This was confirmed by the paired T-test, which displayed significant differences between self-reports and teacher reports (p=0.039).

Informant agreements for the subscales displayed similar patterns. Statistically significant, moderate correlations between parents vs teens, and parents vs teachers (correlations ranged from 0.23 to 0.55). This was confirmed by the paired T-test, where no significant differences were found for any of the subscales between these 2 pairs, except for conduct problems. Self-reports indicated significantly higher levels of conduct problems than did parent reports (p=0.006), and parent reports showed statistically higher levels of conduct problems when compared to teacher reports (p=0.008).

Moreover, low levels of correlations between self-reports and teacher reports remained statistically insignificant for all the subscales, except peer problems (Pearson correlation 0.32, p-value =0.003). This was confirmed by the results of the paired T-test, where statistically significant differences between self-reports and teacher reports were found for all the subscales, except for peer problems.

Table 6: Informant agreement for total difficulty score among parent, teacher, and self-reports

Total Difficulty Score Informant Agreements					
Groups		Mean	Correlation	Sig. †	Paired T-test (p-value) ††
Pair 1 (n=139)	Parent	9.7	0.59**	<0.001	0.164
	Teen	10.3			
Pair 2 (n=86)	Parent	9.1	0.48**	<0.001	0.117
	Teacher	7.9			
Pair 3 (n=86)	Teen	9.8	0.13	0.231	0.039*
	Teacher	7.9			

N= number of subjects

†=Pearson Correlation coefficient, significant results indicate agreement

††= Paired t-test, significant results indicate disagreement

**Pearson correlation significant at level of <0.001 (2-tailed)

* Paired T-test significant at level of 0.05 (2-tailed)

In general, a similar trend was observed across all groups and subscales, where teacher reports showed lower difficulty scores across the groups, followed by parent reports, while self-reports showed the highest scores.

Our third objective was to examine the influence of social factors on the total difficulty scores. Multiple regression analysis was run to examine four possible factors: 1) Number of parents living in the same household as the participant, 2) Number of siblings, 3) Whether the teenager has at least one good friend or not, and 4) Whether they are being bullied or not. The answers to the last 2 questions were collected from the SDQ, based on a scale from 0-2, where 0= Not true, 1= Somewhat true, and 2= Definitely true. We used the parent reports for our regression model, as we assumed them to be the most reliable. The rationale behind this assumption is that parents are expected to understand the overall psychosocial status of their children better than teachers. Also, in comparison to self-reports, parents are expected to report a general long-term evaluation of their child, while self-reports can be influenced by the current emotional status of the affected individual.[110] Our third reason was that parent reports correlated well with both self-reports and teacher reports, while teacher and self-reports showed poor correlations. Thus, we felt that using either self-reports or teacher reports may be less appropriate for this model.

Our regression model showed that the four independent variables statistically significantly predict the total difficulty score ($p=<0.001$), which means

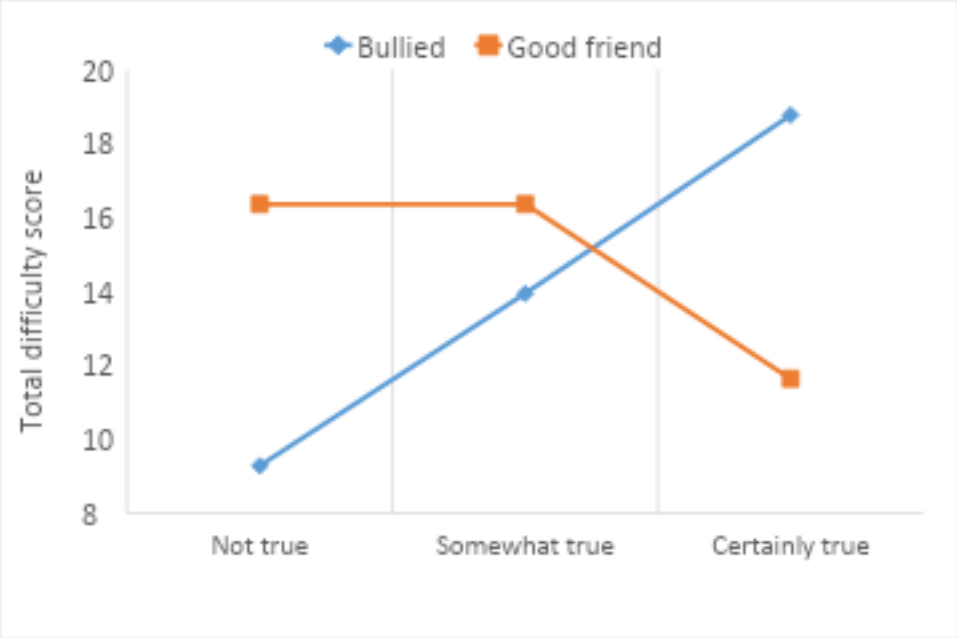
that the regression model is a good fit of the data (Table 7). Moreover, the R^2 value showed that the four factors combined, can explain 44.2% of the total variance of the total difficulty score (Table 8). However, only two of the four factors displayed statistically significant predictions of the total difficulty score (Table 8). The most influential factor was the frequency of being bullied (B= 4.8, p=<0.001), showing that an increase of frequency of being bullied by 1 point, would predict a 4.8-point increase in the total score. This was followed by having a good friend, showing an inverse relationship, where having a good friend would predict a lower total difficulty score (B= -3.5, p=<0.001). Figure 3 shows the association between these 2 significant factors on the total difficulty score. Surprisingly, the number of parents living in the same household was not a significant predictor, although it showed an inverse relationship (B= -1.3, p=0.079), meaning that teenagers living in a 2 parent household, displayed slightly lower difficulties. The poorest predictor was the number of siblings (B= .02, p=0.926), revealing that the number of siblings had almost no impact on the total difficulty score.

Table 7. Regression analysis for the predictive factors

ANOVA	**R square**		
<0.001*	.442		
		B	**Sig.**
Factor	No. of siblings	.024	.926
	No. of parents living in the household	-1.250	.079

	A good friend	-3.526	<.001*
	Frequency of being bullied	4.836	<.001*

Figure 3. Effect of significant social factors on total difficulty score



DISCUSSION

The aim of the current study was to gain a better understanding of the psychosocial functioning of adolescents with CFA and CLP. Our first objective was to examine and compare the psychosocial functioning among adolescents with CFA, CLP and controls. It is of importance to note that for our CON group, the total score and the scores for each subscale were very similar to the normative data for the American population, showing no statistically significant differences. This implies that our control group was appropriately selected, and showing that adolescents

undergoing orthodontic treatment show similar psychosocial functioning compared to the general population.[113,114] In our study, we found no statistically significant differences between males and females, and thus we combined them into one group. This was contrary to previous reports in the literature that found boys to exhibit higher scores, except for emotional problems, compared to girls. [110,111]

Our findings, from all 3 informant groups, show a similar pattern where the CFA group exhibits the highest levels of difficulties, followed by CLP, while the CON group displays the lowest difficulties. Comparing CFA to CON was in accordance with our hypothesis, as parent reports showed significantly higher problems for the total score and all the subscales, except conduct problems and prosocial behavior. Moreover, parent reports showed that adolescents with CFA had statistically significantly higher total difficulties and peer problems, in comparison to CLP. Self-reports and teacher reports showed similar patterns, where all the subscales were higher for CFA, but only the total difficulty score and the peer problems subscale showed statistical significance. In addition, we found no significant differences between CFA and CLP in self-reports or teacher reports.

Although CLP displayed higher levels of difficulties compared to CON on almost all the subscales, they were only statistically significant for peer problems in parent and self-reports, and for the total difficulties in parent reports. However, teacher reports showed no statistically significant differences between CLP and any of the other two groups for any subscale.

Two studies that used different assessment tools reported similar findings among individuals with CFA, as they reported an elevated risk for some psychosocial maladjustments, specifically social problems.[115,116] It is of importance to note that both of these studies had a different design from ours, as they included CLP among other CFA, while we separated them into two different groups. One limitation of our study is that we compiled various CFA into one group, without looking at condition specific responses. Other studies that focused on specific craniofacial conditions found higher levels of behavioral problems among young people with various forms of craniosynostosis and hemifacial macrosomia compared to controls.[55-59,77,117] In contrast, one study found that adjustment levels among individuals with hemifacial microsomia were in level with normative data.[117] We are unaware of any studies that utilized the SDQ to compare adolescents with various CFA, however, some studies used the SDQ to compare individuals with CLP only to those with CLP associated with a wide range of additional conditions. [109,110,111,118] The results of these studies found significantly higher difficulties for subjects with an additional condition besides a cleft, while subjects with only CLP were not significantly different from normative data. [109,110,111,118] Likewise, Brand et al found no statistically significant differences, according to the SDQ, between their CLP group and controls for any subscale.[119] These findings are slightly different from ours, where in our sample of parent reports, we found a significantly higher incidence of total difficulties and peer problems among adolescents with CLP compared to controls. Berger and Dalton

found that mothers of children with CLP reported elevated levels of difficulties on the SDQ, although the children's self-evaluations were within normal levels.[86] This is in agreement with our findings, except that in our sample of self-reports, we found a statistically significant increase in only the peer problems subscale among CLP group. Interestingly, the mean scores for self-reports and parent reports in our CLP sample were very similar to Berger and Dalton's results.[86] We compared our mean SDQ scores of the CLP group to three other studies that utilized the same instrument to evaluate children with CLP in Norway, and found that in our sample, the results of parent reports of the CLP group indicated higher difficulties compared to these three studies.[109,110,111] However, the scores of self-reports of the CLP group were very similar to ours.[109,110,111] This could be explained by the different populations surveyed, since these three studies were conducted in Norway while ours was in the United states. Moreover, their sample was restricted to children aged 10 years old, while ours ranged from 11-17 years old. One possible explanation is a cultural difference between our sample and theirs, where parents in our sample may be more sensitive to psychosocial maladjustments affecting their children. Another possible explanation is the age difference among the samples, as parents of younger children may dismay some problems facing their children, thinking it will resolve as they get older. However, it is worth noting that the sample sizes in those three studies were larger than ours.[109,110,111] While the adjustment levels for adolescents with CLP may lay in the middle between CFA and CON groups, rendering them not statistically

significantly different from either group, it is important to note that results of all three informant groups point towards an increase in psychosocial difficulties levels compared to controls. We recommend that future studies evaluate peer problems in this population in more depth, using specific questionnaires that may uncover some hidden aspects of social interactions.

In our study, we included the extended impact supplement of the SDQ, which to our knowledge was not previously included in other studies in craniofacial research. This supplement evaluates overall distress and impairment and can be summed to generate an impact score. Parents in our CFA sample indicated that their child's psychosocial condition has an adverse impact on their life that averaged 2.5 points on a scale of 0-10. This was statistically significantly higher than the CON group, where the impact averaged only 0.6 points. This is a noteworthy mention, because it sheds the light on how these psychosocial difficulties may disturb various aspects of the adolescent's life.

The SDQ can also be used to classify scores into clinical bandings of normal, borderline, and abnormal, allowing for the identification of individuals above clinical cutoff for adjustment difficulties.[101,104] In our sample, adolescents with CFA exhibited a higher incidence of borderline and abnormal adjustments, followed by CLP, whereas CON group had the lowest incidence of borderline or abnormal behavior (Table 5). These differences were statistically significant for parent and self-reports,

but teacher reports showed no statistically significant differences. The categorical distributions among self-reports and parent reports for the CLP in our study were strikingly similar to those reported by Berger and Dalton, supporting their findings.[86] Perhaps the most alarming finding from our study is that parent reports showed that 34.7% of adolescents with CFA exhibited "abnormal" psychosocial adjustment levels, compared to 17% among CLP and only 3.8% among controls (Table5) (Fig. 2). This 10-fold increase in the risk of developing a psychopathology should be examined in more depth in future studies and supports the need for routine psychosocial screenings of individuals with CFA.

Former investigators have highlighted the importance of using multi-informant studies, in order to obtain a comprehensive, diverse set of data that would allow us to draw more accurate conclusions.[120,121,122] While teacher observations have been widely utilized by social scientists to measure children's psychological adjustment, their presence in the craniofacial literature is scarce, which could be attributed to the practical limitations of gathering these reports. It was interesting to see that in our study, teacher's reports showed the least number of significant differences among the groups. However, they exhibited similar patterns to self-reports or parent reports. The only statistically significant findings from teacher reports were between CFA and CON, for the total difficulties and peer problems only. This could be attributed to the lower participation rate from teachers in our study, which was just over half the number of parent reports. Another consideration is a

possible selection bias since we asked the participants to recommend one teacher, it is likely that they recommended their "favorite" teachers, who they felt most comfortable around. This could have led to a biased response from the teachers, especially when answering questions about their student's behavior. Moreover, teachers may lack knowledge of various aspects of the child's life outside of school, making it harder to evaluate some specific pieces of their student's psychological well-being.

Our second objective was to evaluate the correlations between the three informant groups, to determine how the views of parents and teachers may vary from self-evaluations. Interestingly, we found that across all three groups (CFA, CLP and CON), the correlations between the first pair (parent reports vs self-reports) were moderate, yet significant, for the total score. Similar results were found for the second pair (parent reports vs teacher reports). Interestingly, we found no significant correlations between the third pair (self-reports vs teacher reports) for any of the groups. Since the correlations for all three groups behaved similarly, we combined the data and reported it in one table. (Table 6). As mentioned previously in the results section, the same patterns were observed when comparing the subscales, and thus, we did not feel the need to report them in details. The only exception was a significant correlation between self-reports and teacher reports for the peer problem subscale. A possible explanation for this exception is that the peer problems were the most evident aspect of difficulties, and teachers get to examine this part of a teenager's life

reasonably well during school hours, while other aspects of the teenager's life outside of school may be harder for teachers to evaluate.

In general, the total scores and subscale behaved in a similar pattern, where self-reports showed higher difficulties, followed by parent reports, while teacher reports indicated the lowest scores. It is important to note that poor levels of agreement do not necessarily suggest that the reports are invalid, but rather reflect unique perceptions of each informant.[110,122]

A recent study by Bjerke et al. used the SDQ to evaluate informant agreements for CLP patients and reported a statistically significant difference between self-reports and parent reports for almost all the scales, with children reporting higher levels of difficulties than their parents.[110] The levels of parent-child agreement in their CLP sample were analogous to those found in the normative data.[120] In contrast, our study showed that although self-reports indicated higher levels of difficulties compared to parent reports, there were no statistically significant differences. For all three groups in our sample, higher levels of agreement were found between parent reports and self-reports than what has been reported in the literature. Again, this could be due to cultural differences between the samples, the wider age range that we included, or possibly the relatively modest sample size in our study.

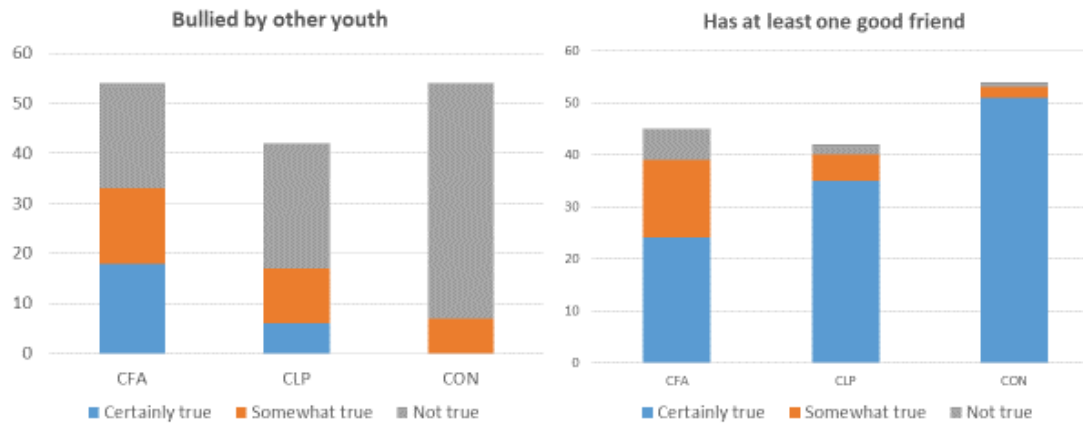
In our study, teacher reports showed lower difficulty scores across the groups, compared to parent or self-reports. One possible reason for this discrepancy is that teachers—in contrast to most parents—offer an adult's view of the child in relation to

a relatively large peer group of the same age, and hence may be more objective in their evaluations, and less sensitive to personal factors. However, it could be due to a lower awareness of the adolescent's personal life outside of school. Another probable reason, as mentioned before, is a possible selection bias of the teachers enrolled. Unfortunately, we found no studies in the craniofacial literature that studied informant agreement among teachers as well, and thus we could not compare our findings to other reports.

Our third objective, was to evaluate four possible predictive social factors for the adolescent's difficulty scores. Our regression analysis showed that the most predictive factor was the frequency of being bullied, followed by having a good friend (Table 7). An increase in the bullying report from "not true" to "certainly true" almost doubles the total difficulty scores (Figure 3). Meanwhile, a negative association can be seen between having a good friend and the total score, where a progress from "not true" to "certainly true" can predict a 5 point decrease on the total difficulty scale (Figure 3). These findings are in agreement with previous studies that supported the notion that social competence is a major factor in mediating psychosocial adjustment levels.[43,44,118] Feragen et al. established an association between social experiences and subjective self-evaluation, suggesting that positive social experiences may preserve self-perceptions of appearance and protect against depressive symptoms.[44] Likewise, Feragen and Stock found an association between social experiences and emotional adjustment levels.[118]

In our sample, 34.1% of teenagers with CFA indicated that they were somewhat bullied, while 18.2% were certainly bullied. These numbers were slightly lower for the CLP subjects (Somewhat true = 26.2%, Certainly true = 14.3%) (Fig. 4). These numbers are particularly disquieting when compared with the control group. Only 13% of our CON group reported being somewhat bullied, while none of the controls answered "certainly bullied". This is in agreement with previous studies that reported elevated levels of teasing and bullying among individuals with CFA.[28,29,30] Moreover, we found it disconcerting that only 53.3% of our CFA subjects indicated that they certainly have a good friend, in comparison to 94.4% of the controls (Fig. 4).

Figure 4. Frequencies of being bullied, and frequencies of having a good friend across the groups



Unexpectedly, we found no significant correlations between the total difficulty scores and the number of parents living in the same household, or the numbers of siblings. Another interesting pattern was observed, where only 60% of the teenagers in the CFA group were living in a two parent household, compared to 75% among the CLP group and 87% among controls, which may support the argument that having children with CFA may complicate parental relationships and be associated with higher rates of divorce. However, although these findings may point to the fact that social interactions outside of home may be more predictive of the adjustment levels than family factors, this should be interpreted with caution. Family interactions are complex and multifaceted, and should be studied in more depth before drawing any definite conclusions.

Our findings support an argument for raising the awareness levels among other adolescents, and a need for the school systems to provide better intervention systems to minimize the prevalence of bullying, with special attention to individuals with CFA. Moreover, we recommend that pediatric psychologists and social workers may consider implementing programs that strengthen the social skills of individuals with CLP or CFA, helping them to integrate themselves better with their peers.

In the current study, efforts were made to overcome some of the aforementioned limitations of previous studies in this field of research. One strength of our study was separating CLP from other CFA, and including an appropriate control group. Our

results indicate that this was appropriate, given the observed trend of increasing difficulties from CON, to CLP, to CFA. Second, we employed teachers' reports, adding a third informant group that is often overlooked in craniofacial research. However, this was not without its own challenges, as the participation rates from teachers ended up being lower than those of parents and subjects, making us more cautious in interpreting teachers' reports. Moreover, we employed a reliable, valid, standardized questionnaire that provides appropriate subscales, and has been widely utilized, allowing us to compare our findings to those of other researchers. Nonetheless, our study had its own limitations as well. One limitation was grouping multiple CFA into one group without analyzing condition specific responses. Future studies may consider multi-center approaches, in order to obtain enough sample sizes to examine specific conditions. A second limitation was not controlling for socio-economic status among the groups, which may be a confounding factor. Other limitations of our study were the limited sample size, and that data were collected at only one time-point, whereas longitudinal studies may provide more comprehensive evaluations of this population. We recommend that future studies should consider studying specific craniofacial conditions separately instead of combining them, employ longitudinal studies, use larger sample sizes and try to evaluate peer problems and social influences in more depth. An alarmingly high incidence of abnormal scores in the CFA group was observed in our study, which may indicate a possible psychopathology. Given the simplicity and brevity of the SDQ, along with its

satisfactory sensitivity and specificity, we think it may be appropriate for orthodontists or craniofacial teams to consider using it for routine psychosocial screening of individuals with CFA.

Conclusions

- A general trend was observed for all informant groups and across all subscales, except prosocial subscale, where CFA group displayed the highest difficulties, followed by CLP, and the lowest were the CON group. These difficulties were specifically emphasized for both CFA and CLP at the peer problems subscale.
- In comparison to controls, adolescents with CLP displayed significantly higher levels of total difficulties according to parent reports, and a significantly higher level of peer problems according to parent and self-reports.
- Informant agreements were moderate, yet significant, for parent reports vs self-reports, and parent reports vs teacher reports. However, no significant correlations were found between self-reports vs teacher reports. A general trend was observed where self-reports indicated the highest difficulty levels, while teacher reports displayed the lowest scores.
- The most predictive factor for the total difficulties score was the frequency of being bullied, followed by having a good friend, highlighting the influence of

social factors. However, the number of parents in the household or the number of siblings did not show significant correlations to the total difficulty score.

- Our findings highlight the need for further studies that explore specific craniofacial conditions separately, with a focus on peer problems and social influences.

MANUSCRIPT REFERENCES

1. Converse JM, Hogan VM, McCarthy JG. Cleft lip and palate. In: Converse JM, editor. Reconstructive Plastic Surgery. 2nd ed. Philadelphia: Saunders; 1977. p. 1930.
2. Savona-Ventura C. The influence of the Roman Catholic Church on Midwifery Practice in Malta. Medical History 1995;39:25-6
3. Dorrance GM. The operative story of cleft palate. Philadelphia: Saunders; 1933.
4. Dunn PM (2006). "Aristotle (384–322 bc): philosopher and scientist of ancient Greece". *Archives of Disease in Childhood: Fetal and Neonatal Edition*. 91: F75–7.
5. Plato., Ferrari G, Griffith T. The republic. Cambridge: Cambridge University Press; 2000.
6. Buchanan, E. P., Xue, A. S., & Hollier, L. H., Jr. (2014). Craniofacial syndromes. Plasticand Reconstructive Surgery, 2014; 134, 128e–153e.
7. Tanaka SA, Mahabir RC, Jupiter DC, Menezes JM. Updating the epidemiology of isolated cleft palate. Plast Reconstr Surg. 2013;131: 650e–652e
8. Pausch, N. C., Winter, K., Halama, D., Wirtz, C., Yildirim, V., & Nattapong, S. Psychosocial acceptance of cleft patients: Has something changed? Journal ofOral and Maxillofacial Surgery, 2006 ;20: 19–26.
9. Partridge, J. Psycho-social reflections on craniofacial morphogenesis. Seminars in Cell and Developmental Biology, 2010;21: 333–338.
10. Olasoji HO, Ugboko VI, Arotiba GT. Cultural and religious components in Nigerian parents' perceptions of the aetiology of cleft lip and palate: implications for treatment and rehabilitation. The British journal of oral & maxillofacial surgery. 2007; 45: 302-305,
11. Johnson KA. China's Hidden Children: Abandonment, Adoption, and the Human Costs of the One-Child Policy. Chicago, IL: University of Chicago Press; 2016.
12. Ryan S., Oaten M., Stevenson R.J., Case T.I. Facial disfigurement is treated like an infectious disease. Evolution and Human Behavior, Vol. 33(6): 639 – 64
13. Roberts RM, Neate GM, Gierasch A. Implicit attitudes towards people with visible difference: findings from an Implicit Association Test. Psychol Health Med. 2017;22(3):352-358

14. Kesande T, Muwazi LM, Bataringaya A, Rwenyonyi CM. Prevalence, pattern and perceptions of cleft lip and cleft palate among children born in two hospitals in Kisoro District, Uganda. BMC Oral Health. 2014; 18:14-104.

15. Adeyemo WL, James O1, Butali A. Cleft lip and palate: Parental experiences of stigma, discrimination, and social/structural inequalities. Ann Maxillofac Surg. 2016;6(2):195-203.

16. Pausch NC, Herzberg PY, Wirtz C, Hemprich A, Dhanuthai K, Hierl T, Pitak-Arnnop P. German animal terms for oral cleft deformity: a Leipzig survey. J Craniomaxillofac Surg, 2012 40:236–242.

17. Goffman, E. Stigma: Notes On the Management of Spoiled Identity. Englewood Cliffs, N.J.: Prentice-Hall. 1963.

18. Langlois, J. H., Kalakanis, L., Rubenstein, A. J., Larson, A., Hallam, M., & Smoot, M. Maxims or myths of beauty? A meta-analytic and theoretical review. Psychological Bulletin, 2000;126(3), 390-423.

19. Kapp K. Self-concept of the cleft lip and or palate child. Cleft Palate J. 1979;16(2):171-6.

20. Topolski TD, Edwards TC, Patrick DL. Quality of life: how do adolescents with facial differences compare with other adolescents. Cleft Palate Craniofac J. 2005;42(1):25-32.

21. Broder H, Strauss RP. Self-concept of early primary school age children with visible or invisible defects. Cleft Palate J. 1989;26(2):114-7.

22. Macgregor FC. Some psychological problems associated with facial deformity. AM Social Rev 1951; 16:629-638.

23. Richman LC. Behavior and achievement of cleft palate children. Cleft Palate J. 1976 Jan;13:4-10.

24. Richman LC, Eliason M. Psychological characteristics of children with cleft lip and palate: intellectual, achievement, behavioral and personality variables. Cleft Palate J. 1982;19(4):249-57.

25. Kapp-Simon K. Self-concept of primary-school-age children with cleft lip, cleft palate, or both. Cleft Palate J. 1986;23(1):24-7.

26. Kapp-Simon KA, McGuire DE. Observed social interaction patterns in adolescents with and without craniofacial conditions. Cleft Palate Craniofac J, 1997 34:380–384

27. N. Rumsey. Body image and congenital conditions with visible differences. T.F. Cash, T. Pruzinsky (Eds.), Body image. A handbook of theory, research, and clinical practice, The Guilford Press, New York, USA (2002), pp. 226-233.

28. Turner, S., Thomas, P., Dowell, T., Rumsey, N., & Sandy, J. Psychological outcomes amongst cleft patients and their families. British Journal of Plastic Surgery, 1997; 50, 1– 9.

29. Lorot-Marchand A., Guerreschi P., Pellerin P., Martinot V., Gbaguidi CC., Neiva C., Devauchelle B., Frochisse C., Poli-Merol ML., Francois-Fiquet C. Frequency and socio-psychological impact of taunting in school-age patients with cleft lip-palate surgical repair. Int J Pediatr Otorhinolaryngol. 2015;79(7):1041-8.

30. Feragen KB, Stock NM. A longitudinal study of 340 young people with or without a visible difference: The impact of teasing on self-perceptions of appearance and depressive symptoms. Body Image. 2016; 16:133-42.

31. Craig W, Harel-Fisch Y, Fogel-Grinvald H, Dostaler S, Hetland J, Simons-Morton B, Molcho M, de Mato MG, Overpeck M, Due P, Pickett W. A cross-national profile of bullying and victimization among adolescents in 40 countries. Int J Public Health. 2009; 54 Suppl 2:216-24.

32. Nansel TR, Overpeck M, Pilla RS, Ruan WJ, Simons-Morton B, Scheidt P. Bullying behaviors among US youth: prevalence and association with psychosocial adjustment. JAMA. 2001;285(16):2094-100.

33. Brunstein Klomek A, Marrocco F, Kleinman M, Schonfeld IS, Gould MS. Bullying, depression, and suicidality in adolescents. J Am Acad Child Adolesc Psychiatry. 2007; 46(1):40-9.

34. Modecki KL, Minchin J, Harbaugh AG, Guerra NG, Runions KC. Bullying prevalence across contexts: a meta-analysis measuring cyber and traditional bullying. J Adolesc Health. 2014;55(5):602-11.

35. Feragen KB, Borge AI. Peer harassment and satisfaction with appearance in children with and without a facial difference. Body Image. 2010;7(2):97-105.

36. Roberts RM, Mathias JL. Psychosocial functioning in adults with congenital craniofacial conditions. Cleft Palate Craniofac J. 2012;49(3):276-85.

37. Danino A, Gradell J, Malka G, Moutel G, Hervé C, Rosilio C. Social adjustment in French adults from who had undergone standardised treatment of complete unilateral cleft lip and palate. Ann Chir Plast Esthet. 2005;50(3):202-5.

38. McWilliams BJ, Paradise LP. Educational, occupational, and marital status of cleft palate adults. Cleft Palate J. 1973; 10:223–229

39. Noar JH. Questionnaire survey of attitudes and concerns of patients with cleft lip and palate and their parents. Cleft Palate Craniofac J. 1991;28:279–284.

40. Sarwer DB, Bartlett SP, Whitaker LA, Paige KT, Pertschuk MJ, Wadden TA. Adult psychological functioning of individuals born with craniofacial anomalies. Plast Reconstr Surg. 1999;103(2):412-8.

41. Madera JM, Hebl MR. Discrimination against facially stigmatized applicants in interviews: an eye-tracking and face-to-face investigation. J Appl Psychol. 2012; 97(2):317–330.

42. Hamlet C, Harcourt D. Older adults' experiences of living with cleft lip and palate: a qualitative study exploring aging and appearance. Cleft Palate Craniofac J. 2015; 52(2):e32-40

43. Berger ZE, Dalton LJ. Coping with a cleft II: Factors associated with psychosocial adjustment of adolescents with a cleft lip and palate and their parents. Cleft Palate Craniofac J. 2011;48(1):82-90

44. Feragen KB, Kvalem IL, Rumsey N, Borge AI. Adolescents with and without a facial difference: The role of friendships and social acceptance in perceptions of appearance and emotional resilience. Body Image. 2010;7(4):271-9.

45. N. Rumsey, N.M. Stock. Living with a cleft: Psychological challenges, support and intervention. S. Berkowitz (Ed.), Cleft lip and palate. Diagnosis and management, Springer Berlin Heidelberg, Chicago, IL, USA (2013), pp. 907-915

46. Strauss RP, Broder H, Helms RW. Perceptions of appearance and speech by adolescent patients with cleft lip and palate and by their parents. Cleft Palate J. 1988;25(4):335-42.

47. Marcusson A, Paulin G, Ostrup L. Facial appearance in adults who had cleft lip and palate treated in childhood. Scand J Plast Reconstr Surg Hand Surg. 2002;36(1):16-23.

48. Heller A, Tidmarsh W, Pless IB. The psychosocial functioning of young adults born with cleft lip or palate. A follow-up study. Clin Pediatr (Phila). 1981;20(7):459-65.

49. Broder H L, Richman L C. An examination of mental health services for cleft and craniofacial patients. Cleft Palate and Craniofacial J. 1987;24: 158-163

50. Broder H, Strauss RP. Psychosocial problems and referrals among oral-facial team patients. Journal of Oral Rehabilitation. 1991;57(1):31-36.

51. Kapp K. Self-concept of the cleft lip and/or palate child. Cleft Palate J 1979; 16:171.

52. Berk N W, Cooper M E, Liu Y E, Marazita M L. Social anxiety in Chinese adults with oral-facial clefts. Cleft Palate-Craniofacial J. 2001;38: 126–133.

53. Nicholls W, Harper C, Selvey LA, Robinson S, Hartig G, Persson M. Body Esteem in a Western Australian Cleft Lip and/or Palate Cohort Across 3 Age Groups. Cleft Palate Craniofac J. 2018;55(4):487-498.

54. Maris CL, Endriga MC, Omnell ML, Speltz ML. Psychosocial adjustment in twin pairs with and without hemifacial microsomia. Cleft Palate Craniofac J. 1999; 36(1):43-50.

55. Broder HL, Smith FB, Strauss RP. Developing a behavior rating scale for comparing teachers' ratings of children with and without craniofacial anomalies. Cleft Palate Craniofac J. 2001;38(6):560-5.

56. Becker DB, Petersen JD, Kane AA, Cradock MM, Pilgram TK, Marsh JL. Speech, cognitive, and behavioral outcomes in nonsyndromic craniosynostosis. Plast Reconstr Surg. 2005;116(2):400-7.

57. Kelleher MO, Murray DJ, McGillivary A, Kamel MH, Allcutt D, Earley MJ. Behavioral, developmental, and educational problems in children with nonsyndromic trigonocephaly. J Neurosurg. 2006;105(5 Suppl):382-4.

58. Bannink N, Maliepaard M, Raat H, Joosten KF, Mathijssen IM. Health-related quality of life in children and adolescents with syndromic craniosynostosis. J Plast Reconstr Aesthet Surg. 2010; 63(12):1972-81.

59. Maliepaard M, Mathijssen IM, Oosterlaan J, Okkerse JM. Intellectual, behavioral, and emotional functioning in children with syndromic craniosynostosis. Pediatrics. 2014 ; 133(6):1608-15.

60. Krueckeberg SM, Kapp-Simon KA. Longitudinal follow-up of social skills in children with and without craniofacial anomalies. Presented at the Annual Meeting of the American Cleft Palate-Craniofacial Association, 1997; New Orleans, LA.

61. Speltz ML, Morton K, Goodell EW, Clarren SK. Psychological functioning of children with craniofacial anomalies and their mothers: follow-up from late infancy to school entry. Cleft Palate J 1993;30:482-489.

62. Wehby GL, Ohsfeldt RL, Murray JC. Health professionals' assessment of health-related quality of life values for oral clefting by age using a visual analogue scale method. Cleft Palate Craniofac J. 2006b; 43:383–91.

63. Kapp-Simon KA, Simon DJ, Kristovich S. Self-perception, social skills, adjustment, and inhibition in young adolescents with craniofacial anomalies. Cleft Palate Craniofac J 1992;29:352–356.

64. Damiano P, Tyler M, Romitti P, Momany E, Jones M, Canady J, Karnell M, Murray J. Health-related quality of life among preadolescent children with oral clefts. Pediatrics. 2007; 120:e283–90.

65. Marcusson A, Akerlind I, Paulin G. Quality of life in adults with repaired complete cleft lip and palate. Cleft Palate Craniofac J. 2001;38:379–385.

66. Endriga MC, Kapp-Simon KA. Psychological issues in craniofacial care: state of the art. Cleft Palate Craniofac J. 1999 ;36(1):3-11.

67. Hutchinson K, Wellman MA, Noe DA, Kahn A. The psychosocial effects of cleft lip and palate in non-Anglo populations: a cross-cultural meta-analysis. Cleft Palate Craniofac J. 2011;48(5):497-508.

68. Nicholls W., Persson M., Robinson S., Selvey L. Psychosocial Impacts of Cleft: A Review of the Literature. AASCIT Journal of Psychology. 2016; 1(2): 14-32.

69. Christensen K, Mortensen PB. Facial clefting and psychiatric diseases: a follow-up of the Danish 1936–1987 Facial Cleft cohort. Cleft Palate Craniofac J. 2002; 39(4):392–6.

70. Demir T, Karacetin G, Baghaki S, Aydin Y. Psychiatric assessment of children with nonsyndromic cleft lip and palate. Gen Hosp Psychiatry. 2011;33(6):594-603.

71. Ramstad T, Ottem E, Shaw WC. Psychosocial adjustment in Norwegian adults who had undergone standardised treatment of complete cleft lip and palate: self-reported problems and concerns with appearance. Scand J Plast Reconstr Hand Surg. 1995b;29:329– 336

72. Nicholls W, Harper C, Robinson S, Persson M, Selvey L. Adult-Specific Life Outcomes of Cleft Lip and Palate in a Western Australian Cohort. Cleft Palate Craniofac J. 2018. *In Press.*

73. Tovetjärn R, Tarnow P, Maltese G, Fischer S, Sahlin PE, Kölby L. Children with Apert syndrome as adults: a follow-up study of 28 Scandinavian patients. Plast Reconstr Surg. 2012;130(4):572e-576e.

74. Christensen K, Juel K, Herskind AM, Murray JC. Long term follow up study of survival associated with cleft lip and palate at birth. BMJ. 2004; 328:1405–1408.

75. Herskind A, Christensen K, Juel K, Fogh-Anderson P. Cleft lip: a risk factor for suicide. Paper presented at: 7th International Congress on Cleft Palate and Related Craniofacial Anomalies; November 2, 1993; Broadbeach, Queensland, Australia.

76. Cloonan YK, Collett B, Speltz ML, Anderka M, Werler MM. Psychosocial outcomes in children with and without non-syndromic craniosynostosis: findings from two studies. Cleft Palate Craniofac J. 2013; 50(4):406-13.

77. Dufton LM, Speltz ML, Kelly JP, Leroux B, Collett BR, Werler MM. Psychosocial outcomes in children with hemifacial microsomia. J Pediatr Psychol. 2011; 36(7):794-805

78. Wogelius P, Gjorup H, Haubek D, Lopez R, Poulsen S. Development of Danish version of child oralhealth-related quality of life questionnaires (CPQ8–10 and CPQ11–14). BMC Oral Health. 2009; 22.9(1):11

79. Oosterkamp BC, Dijkstra PU, Remmelink HJ, Van Oort RP, Goorhuis-Brouwer SM, Sandham A, DeBont LG. Satisfaction with treatment outcome in bilateral cleft lip and palate patients. Int J Oral Maxillofac Surg. 2007; 36:890–5.

80. Aravena PC, Gonzalez T, Oyarzún T, Coronado C. Oral Health-Related Quality of Life in Children in Chile Treated for Cleft Lip and Palate: A Case-Control Approach. Cleft Palate Craniofac J. 2017;54(2):e15-e20

81. Geels LM, Kieffer JM, Hoogstraten J, Prahl-Andersen B. Oral health-related quality of life of children with craniofacial conditions. Cleft Palate Craniofac J. 2008;45(5):461-7.

82. Knudsen E, Maltese G, Tarnow P, Tovetjärn R, Kölby L. Parental estimation of early psychological development in children operated on for single suture synostosis. J Plast Surg Hand Surg. 2012;46(3-4):152-4.

83. Prescott T, Redfors M, Rustad CF, Eiklid KL, Geirdal AØ, Storhaug K, Jensen JL. Characterization of a Norwegian cherubism cohort; molecular genetic findings, oral manifestations and quality of life. Eur J Med Genet. 2013;56(3):131-7

84. Raposo-Amaral CE, Raposo-Amaral CA, Garcia Neto JJ, Farias DB, Somensi RS. Apert syndrome: quality of life and challenges of a management protocol in Brazil. J Craniofac Surg. 2012;23(4):1104-8.

85. Raposo-Amaral CE, Neto JG, Denadai R, Raposo-Amaral CM, Raposo-Amaral CA. Patient-reported quality of life in highest-functioning Apert and Crouzon syndromes: a comparative study. Plast Reconstr Surg. 2014;133(2):182e-91e.

86. Berger ZE, Dalton LJ. Coping with a cleft: psychosocial adjustment of adolescents with a cleft lip and palate and their parents. Cleft Palate Craniofac J. 2009;46(4):435-43

87. Feragen KB, Stock NM, Sharratt ND, Kvalem IL. Self-perceptions of romantic appeal in adolescents with a cleft lip and/or palate. Body Image. 2016;18:143-52.

88. Patel Z, Ross E. Reflections on the cleft experience by South african adults: use of qualitative methodology. Cleft Palate Craniofac J. 2003;40(5):471-80.

89. Eiserman W. Unique outcomes and positive contributions associated with facial difference: expanding research and practice. Cleft Palate Craniofac J. 2001;38(3):236-44.

90. Stock NM, Feragen KB, Rumsey N. Adults' Narratives of Growing up With a Cleft Lip and/or Palate: Factors Associated With Psychological Adjustment. Cleft Palate Craniofac J. 2016;53(2):222-39.

91. Klein T, Pope AW, Getahun E, Thompson J. Mothers' reflections on raising a child with a craniofacial anomaly. Cleft Palate Craniofac J. 2006 Sep;43(5):590-7.

92. Baker SR, Owens J, Stern M, Willmot D. Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. Cleft Palate Craniofac J. 2009;46(3):229-36

93. Kapp-Simon KA, McGuire DE, Long BC, Simon DJ. Addressing quality of life issues in adolescents: social skills interventions. Cleft Palate Craniofac J. 2005;42(1):45-50.

94. Stock NM, Feragen KB, Rumsey N. "It Doesn't All Just Stop at 18": Psychological Adjustment and Support Needs of Adults Born With Cleft Lip and/or Palate. Cleft Palate Craniofac J. 2015;52(5):543-54.

95. Loewenstein J, Sutton E, Guidotti R, Shapiro K, Ball K, McLean D, Biesecker B. The art of coping with a craniofacial difference: helping others through "Positive Exposure". Am J Med Genet A. 2008;146A(12):1547-57

96. Strauss RP, Fenson C. Experiencing the "good life": literary views of craniofacial conditions and quality of life. Cleft Palate Craniofac J. 2005;42(1):14-8.

97. Kelton RW. Facing up to stigma: workplace and personal strategies. Cleft Palate Craniofac J. 2001;38(3):245-7.

98. Feragen KB, Stock NM. Psychological adjustment to craniofacial conditions (excluding oral clefts): A review of the literature. Psychol Health. 2017;32(3):253-288.

99. Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: a systematic review. Eur J Orthod. 2005; 27(3):274–85

100. Wehby GL, Cassell CH. The impact of orofacial clefts on quality of life and healthcare use and costs. Oral Dis. 2010;16:3–10

101. Goodman R (1997) The Strengths and Difficulties Questionnaire: A Research Note. Journal of Child Psychology and Psychiatry, 38, 581-586.

102. Goodman, R. The extended version of the Strengths and Difficulties Questionnaire as a guide to child psychiatric caseness and consequent burden. Journal of Child Psychology and Psychiatry. 1999;40(5),791 – 799.

103. Goodman R, Scott S. Comparing the Strengths and Difficulties Questionnaire and the Child Behavior Checklist: is small beautiful? J Abnorm Child Psychol. 1999;27(1):17-24.

104. Goodman R. Psychometric properties of the strengths and difficulties questionnaire. J Am Acad Child Adolesc Psychiatry. 2001;40(11):1337-45.

105. Goodman R, Ford T, Simmons H, Gatward R, Meltzer H. Using the Strengths and Difficulties Questionnaire (SDQ) to screen for child psychiatric disorders in a community sample. Br J Psychiatry. 2000;177:534-9.

106. Goodman R, Renfrew D, Mullick M. Predicting type of psychiatric disorder from Strengths and Difficulties Questionnaire (SDQ) scores in child mental health clinics in London and Dhaka. Eur Child Adolesc Psychiatry. 2000;9(2):129-34.

107. Warnick, E. M., M. B. Bracken and S. Kasl. "Screening efficiency of the Child Behavior Checklist and Strengths and Difficulties Questionnaire: A systematic review." Child and Adolescent Mental Health, 2008;13(3): 140-147

108. Vostanis P. Strengths and Difficulties Questionnaire: research and clinical applications. Curr Opin Psychiatry. 2006;19:367-372.

109. Feragen KB, Stock NM, Rumsey N. Toward a reconsideration of inclusion and exclusion criteria in cleft lip and palate: implications for psychological research. Cleft Palate Craniofac J. 2014;51(5):569-78

110. Bjerke SM, Feragen KB, Bergvik S. Strengths and Difficulties Questionnaire (SDQ): Informant Agreement Between Children Born With Cleft Lip and/or Palate and Their Parents. Cleft Palate Craniofac J. 2018;55(2):204-212.

111. Feragen KB, Stock NM. When there is more than a cleft: psychological adjustment when a cleft is associated with an additional condition. Cleft Palate Craniofac J. 2014;51(1):5-14.

112. Paul A. Harris, Robert Taylor, Robert Thielke, Jonathon Payne, Nathaniel Gonzalez, Jose G. Conde, Research electronic data capture (REDCap) – A metadata-driven methodology and workflow process for providing translational research informatics support, J Biomed Inform. 2009;42(2):377-81.

113. Bourdon KH, Goodman R, Rae DS, Simpson G, Koretz DS. The Strengths and Difficulties Questionnaire: U.S. normative data and psychometric properties. J Am Acad Child Adolesc Psychiatry. 2005;44(6):557-64.

114. He JP, Burstein M, Schmitz A, Merikangas KR. The Strengths and Difficulties Questionnaire (SDQ): the factor structure and scale validation in U.S. adolescents. J Abnorm Child Psychol. 2013 May;41(4):583-95

115. Pope AW, Snyder HT. Psychosocial adjustment in children and adolescents with a craniofacial anomaly: age and sex patterns. Cleft Palate Craniofac J. 2005;42(4):349-54.

116. Singh VP, Moss TP. Psychological impact of visible differences in patients with congenital craniofacial anomalies. Prog Orthod. 2015;16:5.

117. Snyder H, Pope AW. Psychosocial adjustment in children and adolescents with a craniofacial anomaly: diagnosis-specific patterns. Cleft Palate Craniofac J. 2010;47(3):264-72.

118. Feragen KB, Stock NM. Risk and Protective Factors at Age 10: Psychological Adjustment in Children With a Cleft Lip and/or Palate. Cleft Palate Craniofac J. 2016;53(2):161-79

119. Brand S, Blechschmidt A, Müller A, Sader R, Schwenzer-Zimmerer K, Zeilhofer HF, Holsboer-Trachsler E. Psychosocial functioning and sleep patterns in children and adolescents with cleft lip and palate (CLP) compared with healthy controls. Cleft Palate Craniofac J. 2009;46(2):124-35

120. Van Roy B, Grøholt B, Heyerdahl S, Clench-Aas J. Understanding discrepancies in parent-child reporting of emotional and behavioral problems: effects of relational and socio-demographic factors. BMC Psychiatry. 2010;10:56-68.

121. Offord DR, Boyle MD, Racine Y, Szatmari P, Fleming JE, Sanford M, Lipman EL. Integrating assessment data from multiple informants. J Am Acad Child Adolesc Psychiatry. 1996;35:1078-1085.

122. Achenbach TM, McConaughy SH, Howell CT. Child/adolescent behavioral and emotional problems: implications of crossinformant correlations for situational specificity. Psychol Bull. 1987;101:213-232.

APPENDICES

1. **Table 1.** Number of subjects in each group with age and gender distribution

		CFA	CLP	CON	Total	Sig.†
Number of subjects	Parent reports	49	41	52	142	
	Self-reports	49	40	55	144	
	Teacher reports	26	23	37	86	
	Total	49	42	55	146	
Gender	Male	44.9%	45%	47.3%	43.2 %	**NS**
	Female	55.1%	55%	52.7%	56.8 %	
Mean age		12.5	12.3	13.8	12.9	**NS**

† = One way ANOVA to determine significant differences in the age or gender among the groups

NS= Not statistically significant

2. **Table 2.** Parent reports (n=142) - SDQ scores and subscales

	CFA (n=49)	CLP (n=41)	CON (n=52)	Sig. †	Tukey Post-Hoc		
	Mean ± SD	Mean ± SD	Mean±SD		Group	Mean diff.	Sig.*
Total Difficulties	13.1 ± 6.7	9.8 ± 6.3	6.3 ± 4.6	<0.001**	CFA vs CLP	3.3	0.027 *
					CFA vs CON	6.8	<0.001 **
					CLP vs CON	3.5	0.016*
Emotional problems	3.5 ± 2.4	2.6 ± 2.3	1.8 ± 2.0	<0.001**	CFA vs CON	1.7	<0.001 **
Conduct problems	1.5 ± 1.8	1.4 ± 1.7	0.9 ± 1.2	.115			
Hyperactivity	4.5 ± 2.9	3.6 ± 2.7	2.3 ± 2.4	<0.001**	CFA vs CON	2.2	<0.001 **
Peer Problems	3.5 ± 1.8	2.1 ± 2.1	1.3 ± 1.1	<0.001**	CFA vs CLP	1.4	<0.001 **
					CFA vs CON	2.2	<0.001 **
					CLP vs CON	0.8	0.04*
Prosocial	8.1 ± 1.8	8.4 ± 1.7	8.3 ± 1.7	0.69			
Impact on QOL	2.5 ± 2.8	1.7 ± 2.1	0.6 ± 1.5	<0.001**	CFA vs CON	1.9	<0.001 **

N= number of subjects
† = One way ANOVA to compare the means among the groups
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.001

3. **Table 3.** Self reports (n=144) - SDQ scores and subscales

	CFA (n=49)	CLP (n=40)	CON (n=55)	Sig. (p-value)†	Tukey Post-Hoc for significant findings		
	Mean± SD	Mean± SD	Mean ±SD		Group	Mean difference	Sig.*
Total Difficulties	12.0 ± 6.2	10.6 ± 5.4	8.5 ± 1.7	0.005*	CFA vs CON	3.5	0.004*
Emotional problems	3.4 ± 2.6	3 ± 2.2	2.5 ± 2.1	.101			
Conduct problems	1.8 ± 1.7	1.8 ± 1.5	1.3 ± 1.5	0.142			
Hyperactivity	4.0 ± 2.3	3.7 ± 1.9	3.5 ± 2.2	0.459			
Peer Problems	2.8 ± 1.8	2.3 ± 1.7	1.3 ± 1.2	<0.001**	CFA vs CON	1.4	<0.001**
					CLP vs CON	0.9	0.011*
Prosocial	7.9 ± 2.1	7.8 ± 1.8	8.5 ± 1.7	0.145			
Impact on QOL	1.0 ± 1.7	0.9 ± 1.7	0.4 ± 0.9	0.091			

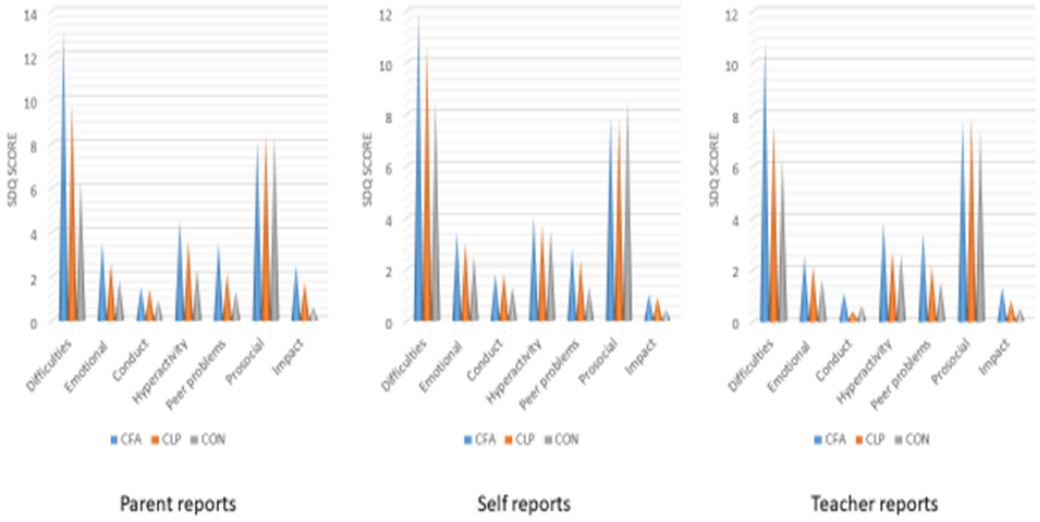
N= number of subjects
† = One way ANOVA to compare the means among the groups
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.001

4. **Table 4.** Teacher reports (n=86) - SDQ scores and subscales

	CFA (n=26)	CLP (n=23)	CON (n=37)	Sig. (p-value)	Tukey Post-Hoc for significant findings		
	Mean± SD	Mean± SD	Mean±SD		Groups	Mean diff.	Sig.*
Total Difficulties	10.8 ±7.9	7.6 ± 5.7	6.3 ± 5.8	0.030 *	CFA vs CON	4.4	0.026 *
Emotional problems	2.5 ± 2.5	2.1 ± 2.0	1.6 ± 2.0	.261			
Conduct problems	1.1 ± 1.7	0.4 ± 0.8	0.6 ± 1.2	0.136			
Hyperactivity	3.8 ± 2.9	2.7 ± 2.7	2.6 ± 3.1	0.288			
Peer Problems	3.4 ± 2.6	2.1 ± 2.4	1.5 ± 1.7	0.004 **	CFA vs CON	1.9	0.003 **
Prosocial	7.7 ± 2.3	7.9 ± 2.5	7.4 ± 2.1	0.680			
Impact on QOL	1.3 ± 1.7	0.8 ± 1.5	0.5 ± 1.1	0.098			

N= number of subjects
† = One way ANOVA to determine significant differences among the groups
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.01

5. **Figure 1.** Mean SDQ scores for all groups.



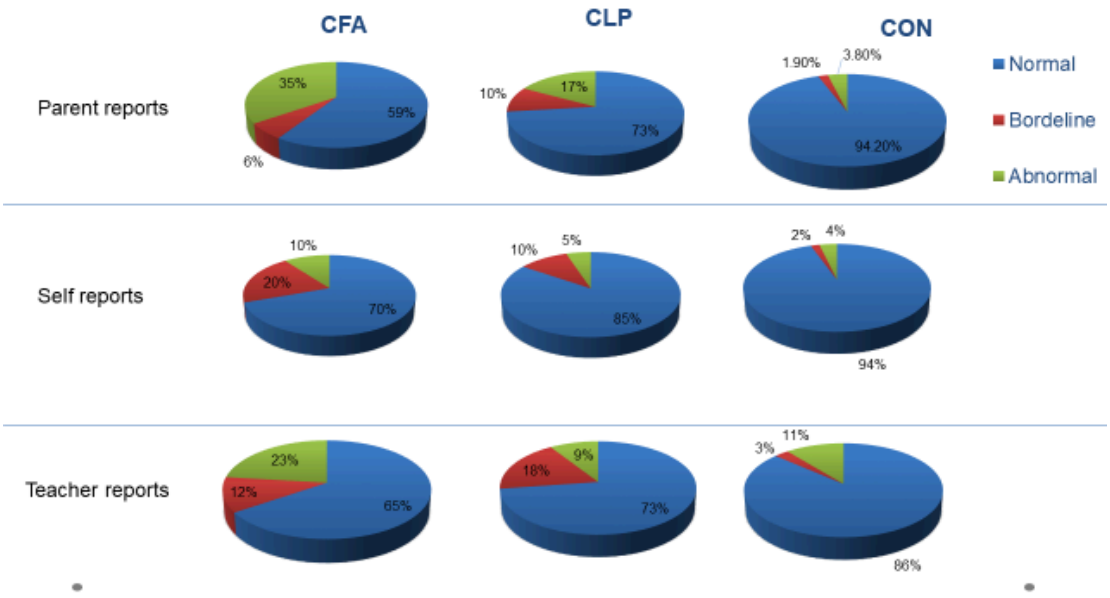
A similar trend is observed across all three informant groups, for all subscales (except prosocial), where the highest levels of difficulties can be observed for the CFA group, followed by CLP, while CON group has the lowest scores.

6. **Table 5.** Frequencies of normal, borderline, and abnormal SDQ scores.

Informant	Category		Group			Total	Sig†
			CFA	CLP	CON		
Parent Reports	Normal	Count	29	30	49	108	**<0.001****
		% in Group	59.2%	73.2%	94.2%	76.1%	
	Borderline	Count	3	4	1	8	
		% in Group	6.1%	9.8%	1.9%	5.6%	
	Abnormal	Count	17	7	2	26	
		% in Group	34.7%	17.1%	3.8%	18.3%	
Self Reports	Normal	Count	34	33	52	119	**0.015***
		% in Group	69.4%	84.6%	94.5%	83.2%	
	Borderline	Count	10	4	1	15	
		% in Group	20.4%	10.3%	1.8%	10.5%	
	Abnormal	Count	5	2	2	9	
		% in Group	10.2%	5.1%	3.6%	6.3%	
Teacher Reports	Normal	Count	17	16	32	65	**0.147**
		% in Group	65.4%	72.7%	86.5%	76.5%	
	Borderline	Count	3	4	1	8	
		% in Group	11.5%	18.2%	2.7%	9.4%	
	Abnormal	Count	6	2	4	12	
		% in Group	23.1%	9.1%	10.8%	14.1%	

† = Chi-Square test for categorical comparisons
*= Statistically significant at a p-value of <0.05
**= Statistically significant at a p-value of <0.001

7. **Figure 2.** Categorical distributions among the groups



8. **Table 6.** Informant agreement among parent, teacher, and self-reports

Total Difficulty Score Informant Agreements					
Groups		Mean	Correlation	Sig.†	Paired T-test††
Pair 1 (n=139)	Parent	9.7	0.59**	<0.001	0.164
	Teen	10.3			
Pair 2 (n=86)	Parent	9.1	0.48**	<0.001	0.117
	Teacher	7.9			
Pair 3 (n=86)	Teen	9.8	0.13	0.231	0.039*
	Teacher	7.9			

N= number of subjects

Pearson Correlation coefficient, significant results indicate agreement at a p-level of <0.001

†† = Paired t-test, significant results indicate disagreement, at a p-level of 0.05 (2-tailed)

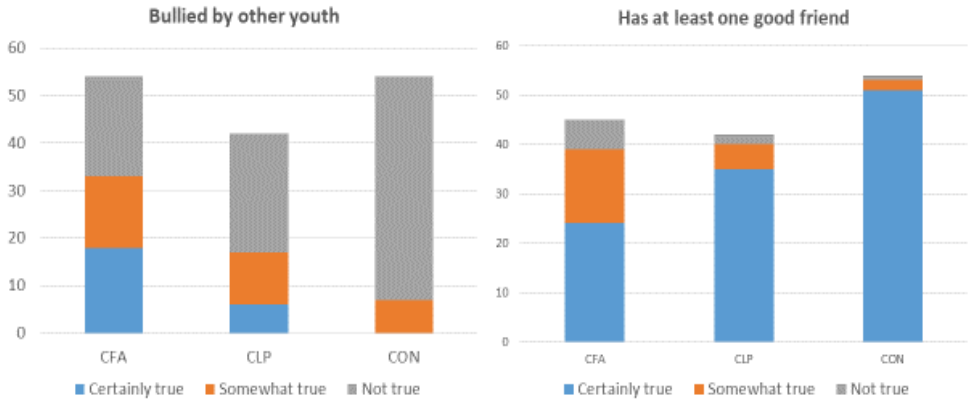
9. **Table 7.** Regression analysis for the predictive factors

ANOVA	**R square**		
<0.001*	.442		
		B	**Sig.**
Factor	No. of siblings	.024	.926
	No. of parents living in the Household	-1.250	.079
	A good friend	-3.526	<.001*
	Frequency of being bullied	4.836	<.001*

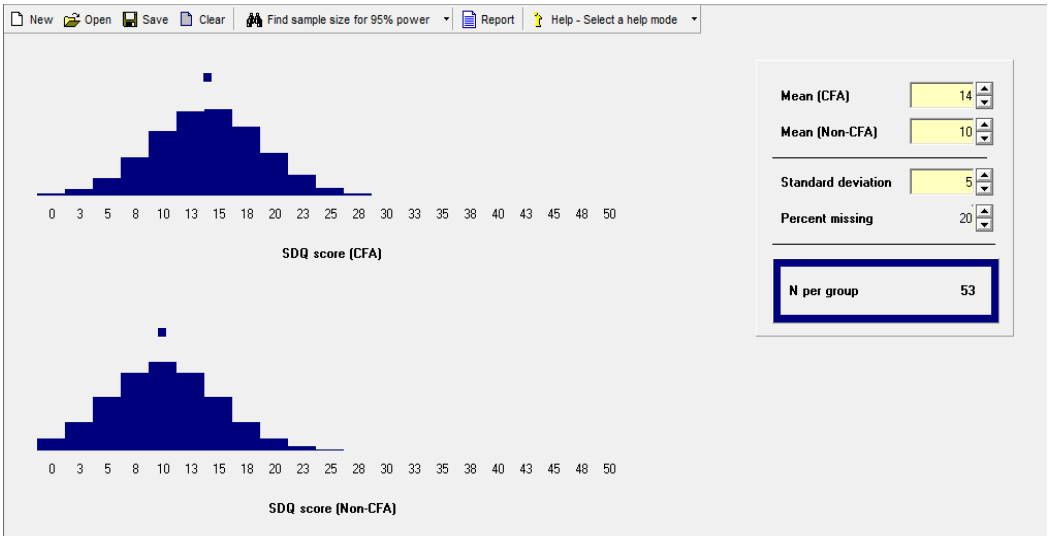
10. **Figure 3.** Effect of significant social factors on total difficulty scor



11. **Figure 4.** Frequencies of being bullied, and frequencies of having a good friend across the groups



12. **Figure 5.** Sample size calculation.



With a sample of 53 subjects/per group, and an assumption that 20% will be missing, we will have a power of 95%. This means that a sample size of 42 (53-20% = 53x0.8), would be appropriate. This level was achieved in our study, since our sample sizes for each group ranged from 42-55.

13. **Figure 6**. Parent version of the SDQ.

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	☐	☐	☐
Restless, overactive, cannot stay still for long	☐	☐	☐
Often complains of headaches, stomach-aches or sickness	☐	☐	☐
Shares readily with other youth, for example books, games, food	☐	☐	☐
Often loses temper	☐	☐	☐
Would rather be alone than with other youth	☐	☐	☐
Generally well behaved, usually does what adults request	☐	☐	☐
Many worries or often seems worried	☐	☐	☐
Helpful if someone is hurt, upset or feeling ill	☐	☐	☐
Constantly fidgeting or squirming	☐	☐	☐
Has at least one good friend	☐	☐	☐
Often fights with other youth or bullies them	☐	☐	☐
Often unhappy, depressed or tearful	☐	☐	☐
Generally liked by other youth	☐	☐	☐
Easily distracted, concentration wanders	☐	☐	☐
Nervous in new situations, easily loses confidence	☐	☐	☐
Kind to younger children	☐	☐	☐
Often lies or cheats	☐	☐	☐
Picked on or bullied by other youth	☐	☐	☐
Often offers to help others (parents, teachers, children)	☐	☐	☐
Thinks things out before acting	☐	☐	☐
Steals from home, school or elsewhere	☐	☐	☐
Gets along better with adults than with other youth	☐	☐	☐
Many fears, easily scared	☐	☐	☐
Good attention span, sees work through to the end	☐	☐	☐

Score ranges from 0 to 40 for difficulties and 0-10 for pro-social. “Somewhat True” is always scored as 1. “Not True: scores 0 for difficulties items, or 2 for pro-social items.
“Certainly True: scores 2 for difficulties, or 0 for pro-social. Higher scores indicate greater difficulties.

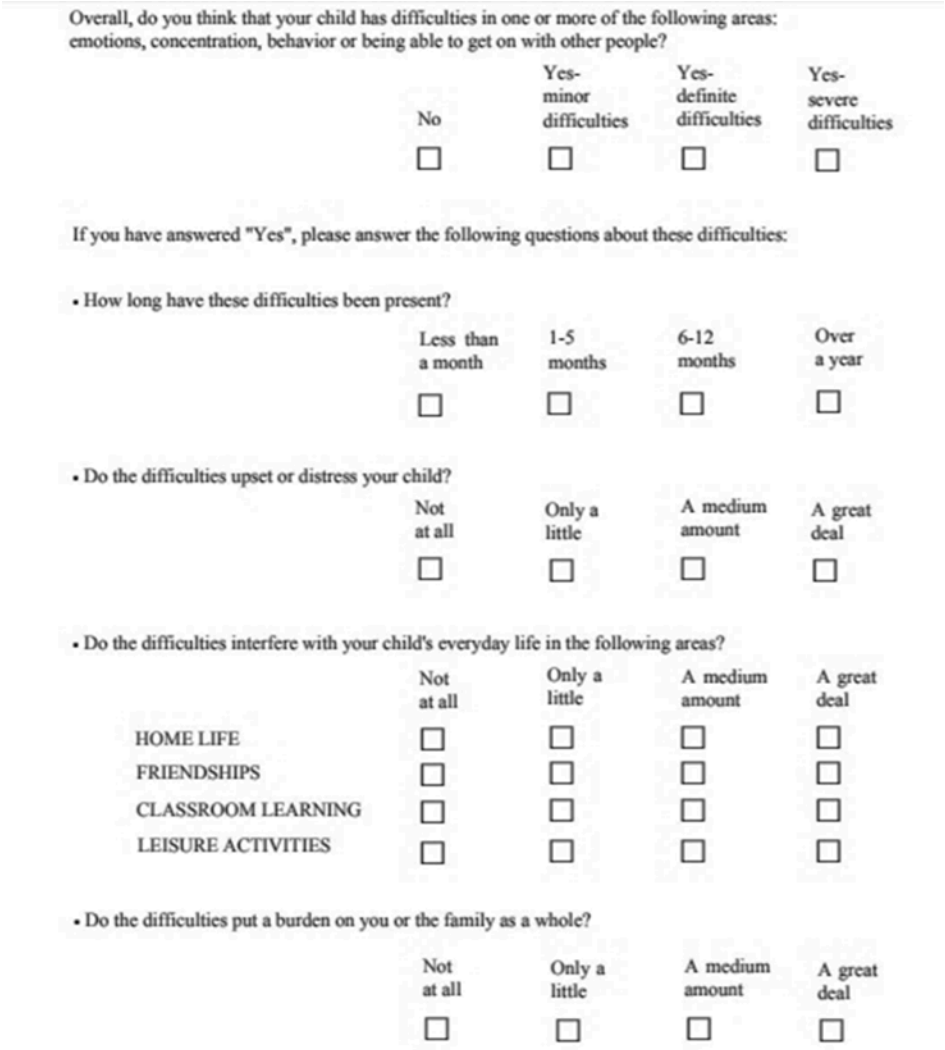
14. **Figure 7.** Self-report version of the SDQ.

	Not True	Somewhat True	Certainly True
I try to be nice to other people. I care about their feelings	☐	☐	☐
I am restless, I cannot stay still for long	☐	☐	☐
I get a lot of headaches, stomach-aches or sickness	☐	☐	☐
I usually share with others, for example CD's, games, food	☐	☐	☐
I get very angry and often lose my temper	☐	☐	☐
I would rather be alone than with people of my age	☐	☐	☐
I usually do as I am told	☐	☐	☐
I worry a lot	☐	☐	☐
I am helpful if someone is hurt, upset or feeling ill	☐	☐	☐
I am constantly fidgeting or squirming	☐	☐	☐
I have one good friend or more	☐	☐	☐
I fight a lot. I can make other people do what I want	☐	☐	☐
I am often unhappy, depressed or tearful	☐	☐	☐
Other people my age generally like me	☐	☐	☐
I am easily distracted, I find it difficult to concentrate	☐	☐	☐
I am nervous in new situations. I easily lose confidence	☐	☐	☐
I am kind to younger children	☐	☐	☐
I am often accused of lying or cheating	☐	☐	☐
Other children or young people pick on me or bully me	☐	☐	☐
I often offer to help others (parents, teachers, children)	☐	☐	☐
I think before I do things	☐	☐	☐
I take things that are not mine from home, school or elsewhere	☐	☐	☐
I get along better with adults than with people my own age	☐	☐	☐
I have many fears, I am easily scared	☐	☐	☐
I finish the work I'm doing. My attention is good	☐	☐	☐

15. **Figure 8.** Teacher version of the SDQ

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	☐	☐	☐
Restless, overactive, cannot stay still for long	☐	☐	☐
Often complains of headaches, stomach-aches or sickness	☐	☐	☐
Shares readily with other youth, for example pencils, books, food	☐	☐	☐
Often loses temper	☐	☐	☐
Would rather be alone than with other youth	☐	☐	☐
Generally well behaved, usually does what adults request	☐	☐	☐
Many worries or often seems worried	☐	☐	☐
Helpful if someone is hurt, upset or feeling ill	☐	☐	☐
Constantly fidgeting or squirming	☐	☐	☐
Has at least one good friend	☐	☐	☐
Often fights with other youth or bullies them	☐	☐	☐
Often unhappy, depressed or tearful	☐	☐	☐
Generally liked by other youth	☐	☐	☐
Easily distracted, concentration wanders	☐	☐	☐
Nervous in new situations, easily loses confidence	☐	☐	☐
Kind to younger children	☐	☐	☐
Often lies or cheats	☐	☐	☐
Picked on or bullied by other youth	☐	☐	☐
Often offers to help others (parents, teachers, children)	☐	☐	☐
Thinks things out before acting	☐	☐	☐
Steals from home, school or elsewhere	☐	☐	☐
Gets along better with adults than with other youth	☐	☐	☐
Many fears, easily scared	☐	☐	☐
Good attention span, sees work through to the end	☐	☐	☐

16. **Figure 9.** The parent version impact supplement of the SDQ.



Overall, do you think that your child has difficulties in one or more of the following areas: emotions, concentration, behavior or being able to get on with other people?

No	Yes-minor difficulties	Yes-definite difficulties	Yes-severe difficulties
☐	☐	☐	☐

If you have answered "Yes", please answer the following questions about these difficulties:

- How long have these difficulties been present?

Less than a month	1-5 months	6-12 months	Over a year
☐	☐	☐	☐

- Do the difficulties upset or distress your child?

Not at all	Only a little	A medium amount	A great deal
☐	☐	☐	☐

- Do the difficulties interfere with your child's everyday life in the following areas?

	Not at all	Only a little	A medium amount	A great deal
HOME LIFE	☐	☐	☐	☐
FRIENDSHIPS	☐	☐	☐	☐
CLASSROOM LEARNING	☐	☐	☐	☐
LEISURE ACTIVITIES	☐	☐	☐	☐

- Do the difficulties put a burden on you or the family as a whole?

Not at all	Only a little	A medium amount	A great deal
☐	☐	☐	☐

The impact supplement identifies the psychological impact on different aspects of the adolescent's life. Score ranges from 0-10 for parents and self-filled reports, and 0-6 for teacher reports.

17. Figure 10. Impact supplement for the self-report version of the SDQ.

Overall, do you think that you have difficulties in any of the following areas: emotions, concentration, behavior or being able to get on with other people?

No	Yes- minor difficulties	Yes- definite difficulties	Yes- severe difficulties
☐	☐	☐	☐

If you have answered "Yes", please answer the following questions about these difficulties:

- How long have these difficulties been present?

Less than a month	1-5 months	6-12 months	Over a year
☐	☐	☐	☐

- Do the difficulties upset or distress you?

Not at all	Only a little	A medium amount	A great deal
☐	☐	☐	☐

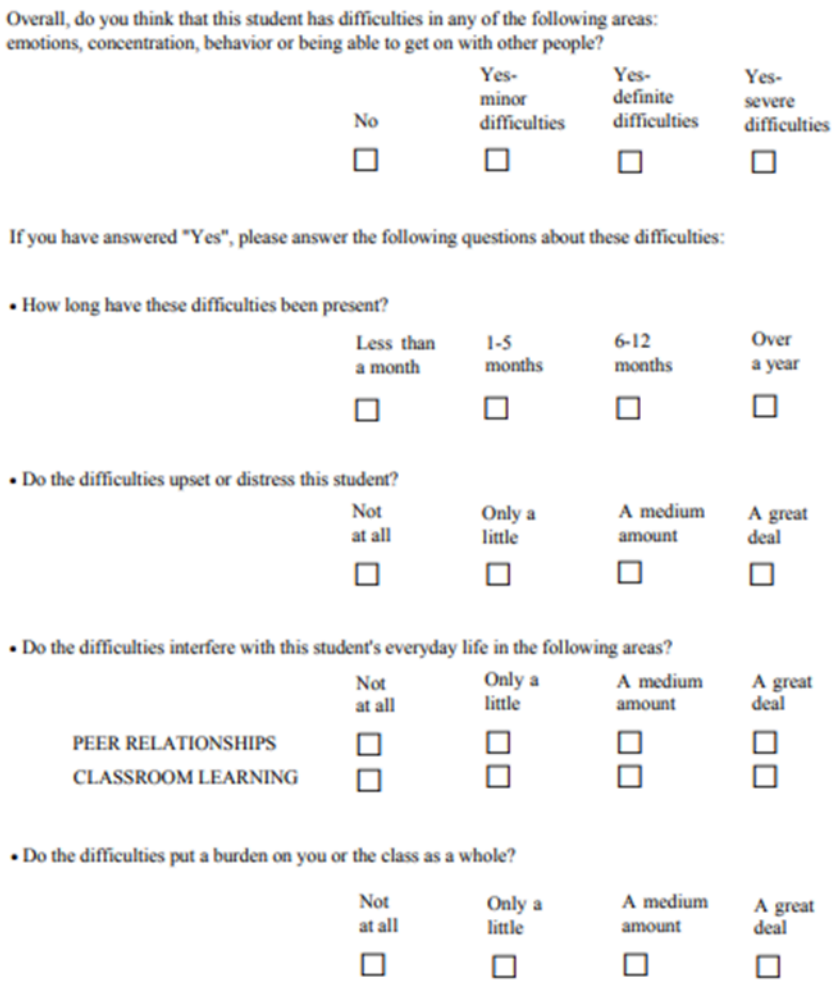
- Do the difficulties interfere with your everyday life in the following areas?

	Not at all	Only a little	A medium amount	A great deal
HOME LIFE	☐	☐	☐	☐
FRIENDSHIPS	☐	☐	☐	☐
CLASSROOM LEARNING	☐	☐	☐	☐
LEISURE ACTIVITIES	☐	☐	☐	☐

- Do the difficulties make it harder for those around you (family, friends, teachers, etc.)?

Not at all	Only a little	A medium amount	A great deal
☐	☐	☐	☐

18. **Figure 11.** Impact supplement for the teacher version of the SDQ.



Overall, do you think that this student has difficulties in any of the following areas: emotions, concentration, behavior or being able to get on with other people?

No	Yes- minor difficulties	Yes- definite difficulties	Yes- severe difficulties
☐	☐	☐	☐

If you have answered "Yes", please answer the following questions about these difficulties:

- How long have these difficulties been present?

Less than a month	1-5 months	6-12 months	Over a year
☐	☐	☐	☐

- Do the difficulties upset or distress this student?

Not at all	Only a little	A medium amount	A great deal
☐	☐	☐	☐

- Do the difficulties interfere with this student's everyday life in the following areas?

	Not at all	Only a little	A medium amount	A great deal
PEER RELATIONSHIPS	☐	☐	☐	☐
CLASSROOM LEARNING	☐	☐	☐	☐

- Do the difficulties put a burden on you or the class as a whole?

Not at all	Only a little	A medium amount	A great deal
☐	☐	☐	☐

19. **Figure 12.** Scoring the SDQ

Scoring the Strengths & Difficulties Questionnaire for age 4-17

The 25 items in the SDQ comprise 5 scales of 5 items each. It is usually easiest to score all 5 scales first before working out the total difficulties score. 'Somewhat True' is always scored as 1, but the scoring of 'Not True' and 'Certainly True' varies with the item, as shown below scale by scale. For each of the 5 scales the score can range from 0 to 10 if all items were completed. These scores can be scaled up pro-rata if at least 3 items were completed, e.g. a score of 4 based on 3 completed items can be scaled up to a score of 7 (6.67 rounded up) for 5 items.

Table 1: Scoring symptom scores on the SDQ for 4-17 year olds

	Not True	Somewhat True	Certainly True
Emotional problems scale			
ITEM 3: Often complains of headaches… *(I get a lot of headaches…)*	0	1	**2**
ITEM 8: Many worries… *(I worry a lot)*	0	1	**2**
ITEM 13: Often unhappy, downhearted… *(I am often unhappy….)*	0	1	**2**
ITEM 16: Nervous or clingy in new situations… *(I am nervous in new situations…)*	0	1	**2**
ITEM 24: Many fears, easily scared *(I have many fears…)*	0	1	**2**
Conduct problems Scale			
ITEM 5: Often has temper tantrums or hot tempers *(I get very angry)*	0	1	**2**
ITEM 7: Generally obedient… *(I usually do as I am told)*	**2**	1	0
ITEM 12: Often fights with other children… *(I fight a lot)*	0	1	**2**
ITEM 18: Often lies or cheats *(I am often accused of lying or cheating)*	0	1	**2**
ITEM 22: Steals from home, school or elsewhere *(I take things that are not mine)*	0	1	**2**
Hyperactivity scale			
ITEM 2: Restless, overactive… *(I am restless…)*	0	1	**2**
ITEM 10: Constantly fidgeting or squirming *(I am constantly fidgeting….)*	0	1	**2**
ITEM 15: Easily distracted, concentration wanders *(I am easily distracted)*	0	1	**2**
ITEM 21: Thinks things out before acting *(I think before I do things)*	**2**	1	0
ITEM 25: Sees tasks through to the end… *(I finish the work I am doing)*	**2**	1	0
Peer problems scale			
ITEM 6: Rather solitary, tends to play alone *(I am usually on my own)*	0	1	**2**
ITEM 11: Has at least one good friend *(I have one goof friend or more)*	**2**	1	0
ITEM 14: Generally liked by other children *(Other people my age generally like me)*	**2**	1	0
ITEM 19: Picked on or bullied by other children… *(Other children or young people pick on me)*	0	1	**2**
ITEM 23: Gets on better with adults than with other children *(I get on better with adults than with people my age)*	0	1	**2**
Prosocial scale			
ITEM 1: Considerate of other people's feelings *(I try to be nice to other people)*	0	1	**2**
ITEM 4: Shares readily with other children… *(I usually share with others)*	0	1	**2**
ITEM 9: Helpful if someone is hurt… *(I am helpful is someone is hurt…)*	0	1	**2**
ITEM 17: Kind to younger children *(I am kind to younger children)*	0	1	**2**
ITEM 20: Often volunteers to help others… *(I often volunteer to help others)*	0	1	**2**

Total difficulties score: This is generated by summing scores from all the scales except the prosocial scale. The resultant score ranges from 0 to 40, and is counted as missing of one of the 4 component scores is missing.

'Externalising' and 'internalising' scores: The externalising score ranges from 0 to 20 and is the sum of the conduct and hyperactivity scales. The internalising score ranges from 0 to 20 and is the sum of the emotional and peer problems scales. Using these two amalgamated scales may be preferable to using the four separate scales in community samples, whereas using the four separate scales may add more value in high-risk samples *(see Goodman & Goodman. 2009 Strengths and difficulties questionnaire as a dimensional measure of child mental health. J Am Acad Child Adolesc Psychiatry 48(4), 400-403).*

Generating impact scores

When using a version of the SDQ that includes an 'impact supplement', the items on overall distress and impairment can be summed to generate an impact score that ranges from 0 to 10 for parent- and self-report, and from 0 to 6 for teacher-report.

Table 2: Scoring the SDQ impact supplement

	Not at all	Only a little	A medium amount	A great deal
Parent report:				
Difficulties upset or distress child	0	0	1	2
Interfere with HOME LIFE	0	0	1	2
Interfere with FRIENDSHIPS	0	0	1	2
Interfere with CLASSROOM LEARNING	0	0	1	2
Interfere with LEISURE ACTIVITIES	0	0	1	2
Teacher report:				
Difficulties upset or distress child	0	0	1	2
Interfere with PEER RELATIONS	0	0	1	2
Interfere with CLASSROOM LEARNING	0	0	1	2
Self-report report:				
Difficulties upset or distress child	0	0	1	2
Interfere with HOME LIFE	0	0	1	2
Interfere with FRIENDSHIPS	0	0	1	2
Interfere with CLASSROOM LEARNING	0	0	1	2
Interfere with LEISURE ACTIVITIES	0	0	1	2

Responses to the questions on chronicity and burden to others are not included in the impact score. When respondents have answered 'no' to the first question on the impact supplement (i.e. when they do not perceive themselves as having any emotional or behavioural difficulties), they are not asked to complete the questions on resultant distress or impairment; the impact score is automatically scored zero in these circumstances.

Cut-points for SDQ scores: original three-band solution and newer four-band solution

Although SDQ scores can be used as continuous variables, it is sometimes convenient to categorise scores. The initial bandings presented for the SDQ scores were 'normal', 'borderline' and 'abnormal'. These bandings were defined based on a population-based UK survey, attempting to choose cutpoints such that 80% of children scored 'normal', 10% 'borderline' and 10% 'abnormal'.

More recently a four-fold classification has been created based on an even larger UK community sample. This four-fold classification differs from the original in that it (1) divided the top 'abnormal' category into two groups, each containing around 5% of the population, (2) renamed the four categories (80% 'close to average', 10% 'slightly raised, 5% 'high' and 5% 'very high' for all scales except prosocial, which is 80% 'close to average', 10% 'slightly lowered', 5% 'low' and 5% 'very low'), and (3) changed the cut-points for some scales, to better reflect the proportion of children in each category in the larger dataset.

Table 3: Categorising SDQ scores for 4-17 year olds

	Original three-band categorisation			Newer four-band categorisation			
	Normal	Borderline	Abnormal	Close to average	Slightly raised (/slightly lowered)	High (/Low)	Very high (very low)
Parent completed SDQ							
Total difficulties score	0-13	14-16	17-40	0-13	14-16	17-19	20-40
Emotional problems score	0-3	4	5-10	0-3	4	5-6	7-10
Conduct problems score	0-2	3	4-10	0-2	3	4-5	6-10
Hyperactivity score	0-5	6	7-10	0-5	6-7	8	9-10
Peer problems score	0-2	3	4-10	0-2	3	4	5-10
Prosocial score	6-10	5	0-4	8-10	7	6	0-5
Impact score	0	1	2-10	0	1	2	3-10
Teacher completed SDQ							
Total difficulties score	0-11	12-15	16-40	0-11	12-15	16-18	19-40
Emotional problems score	0-4	5	6-10	0-3	4	5	6-10
Conduct problems score	0-2	3	4-10	0-2	3	4	5-10
Hyperactivity score	0-5	6	7-10	0-5	6-7	8	9-10
Peer problems score	0-3	4	5-10	0-2	3-4	5	6-10
Prosocial score	6-10	5	0-4	6-10	5	4	0-3
Impact score	0	1	2-10	0	1	2	3-10
Self-completed SDQ							
Total difficulties score	0-15	16-19	20-40	0-14	15-17	18-19	20-40
Emotional problems score	0-5	6	7-10	0-4	5	6	7-10
Conduct problems score	0-3	4	5-10	0-3	4	5	6-10
Hyperactivity score	0-5	6	7-10	0-5	6	7	8-10
Peer problems score	0-3	4-5	6-10	0-2	3	4	5-10
Prosocial score	6-10	5	0-4	7-10	6	5	0-4
Impact score	0	1	2-10	0	1	2	3-10

Note that both these systems only provide a rough-and-ready way of screening for disorders; combining information from SDQ symptom and impact scores from multiple informants is better, but still far from perfect.

20. UHCMC IRB Approval

University Hospitals Cleveland Medical Center

IRB ADMINISTRATION OFFICE
11100 Euclid Avenue, Lakeside 1400
Cleveland, Ohio 44106
Phone: (216) 844-1529

IRB APPROVAL NOTIFICATION

The University Hospitals Institutional Review Board (IRB) has reviewed the following submission:

Principal Investigator: Rany M Bous
Protocol Title: Social-Emotional Coping in Adolescents with Craniofacial Anomalies
UHCMC IRB number: 07-17-39

Submission Type: Addendum
Other, explain:Changes to the E-mail format
Review Type: Expedite

As such, the UHCMC IRB has determined that with respect to the rights and welfare of the individuals, the appropriateness of the methods used to obtain informed consent and the risks and potential medical benefits of the investigation, the current submission is acceptable under Federal Human Subject Protection regulations promulgated under 45 CFR 46 and 21 CFR 50 and 56.

Date of Approval: 12/20/2017

The current expiration date for this study is: 12/05/2018
(The expiration date is the last day that a protocol has IRB approval)

- Per Federal regulation, changes MAY NOT be made to any element of the current research without prior IRB approval, except to eliminate an immediate and apparent hazard to subjects enrolled in the trial.
- Per Federal regulation, the research may not continue without IRB approval. You must submit a request for continuation at least 6-8 weeks prior to the expiration date noted above. Once the study is complete, the IRB requires prompt notification of study closure.
- Failure to retain current IRB approval may result in archiving of the current study and human subjects non-compliance allegations.

Documents reviewed and/or approved as part of this submission:

Title	Version Number	Version Date
Revised enrollment E-mail	Version 1.0	12/17/2017

Human Risk Determination: Not Greater Than Minimal Risk

If you have any questions or concerns, please contact the UHCMC IRB office at (216) 844-1529. The UHCMC IRB appreciates your commitment towards the ethical conduct of human subjects research.

21. Parent Consent Form

07/21/2017

IRB NUMBER: 07-17-39
IRB APPROVAL DATE: 12/06/2017
IRB EXPIRATION DATE: 12/05/2018

Informed Consent

Research Statement

We are currently conducting a study entitled ***"Social-emotional Coping in Adolescents with Craniofacial Conditions"*** that compares the psycho-social adjustment of adolescents with craniofacial conditions to that of adolescents without craniofacial conditions. The following questionnaire is collected for the purpose of research. We are requesting your participation in this study because you are a parent of a teen with a craniofacial condition, who was seen by our craniofacial team.

The main purpose of this study is to identify and compare the psycho-social aspects of adolescents with craniofacial conditions compared to adolescents without craniofacial conditions. Our secondary objectives are to identify support systems that might help them cope better.

The questionnaire takes around 15 minutes to complete. It includes questions about social and behavioral functioning of adolescents. Additional questions involve the support structures, and social-economic status of the families. If you accept to be a part of our research, we will ask you to fill the questionnaire, and have your adolescent son\daughter fill a different version of it as well. Also, we would request that you recommend to us one teacher that you think can fill the teacher version of the questionnaire on behalf of your child. If you do not wish to have a teacher participate, it will not affect your enrollment in the study and you can still participate in the study.

Teachers offer an adult's view of the child in relation to a relatively large peer group of the same age. Also, social encounters outside home can be more difficult for the affected teens, and thus we believe that teacher reports can be helpful in identifying the behavioral functioning of adolescents.

If you and your child accept and recommend a teacher, we will contact the teacher, and explain to them the purpose of our study, and have them fill out the teacher version of the questionnaire. We will collect our data on a secure web portal, to protect the privacy of your information. However, the teacher will preserve the right to deny participation. In that case, your enrollment will not be affected.
If you do not wish for us to contact one your child's teachers, you can still participate in the study.

Risks and discomforts

There is no risk of physical harm to being in this study, but there is a small risk of loss of privacy and confidentiality which we are protecting against by careful handling of all information. We will use secure web portals for the data collection and storage to protect the privacy of our information.

Benefits

Although there are no direct benefits to our participants, we are anticipating a significant benefit to the community, especially those affected with craniofacial conditions or cleft lip and palate. Our findings would allow us to recommend better support systems for schools and parents and help them implement prevention initiatives, with the long-range goal of developing evidence-based intervention strategies by school personnel and pediatric psychologists.

Confidentiality

Efforts will be made to keep the personal information in your research record private and confidential, but absolute confidentiality cannot be guaranteed. The University Hospitals Cleveland Medical Center Institutional Review Board may review your study records. The UH REDCap will be used for data collection and storage. It is a secure web application for building and managing online surveys and databases, that serves to protect the confidentiality and privacy of your information.

Compensation

Each participating teenager will get a $10 Amazon gift card each, as a token of appreciation for being a part of our research.

Contact Person

If you have any questions regarding the study, concerns or complaints about the study in the future, please contact the primary investigator, Dr. Rany Bous. E-mail: rmb184@case.edu. Cell: 440-832-9405.

If the researchers cannot be reached, or if you would like to talk to someone other than the researcher(s) about; concerns regarding the study; research participant's rights; research-related injury; or other human subject issues, please call the University Hospitals Cleveland

Medical Center's Research Subject Rights phone line at (216) 983-4979 or write to: The Associate Chief Scientific Officer, The Clinical Research Center, University Hospitals Cleveland Medical Center, 11100 Euclid Avenue, Lakeside 1400, Cleveland, Ohio, 44106-7061.

Voluntary Participation and Right to Withdraw

Participation is voluntary, refusal to participate will involve no penalty or loss of benefits to which the participant is otherwise entitled and the participant may discontinue participation at any time.

Thank you for taking the time to read this, your participation is highly appreciated.

22. Child Assent form



IRB NUMBER: 07-17-39
IRB APPROVAL DATE: 12/06/
IRB EXPIRATION DATE: 12/05

We are conducting a study at the University Hospitals Craniofacial Team/Case Western Reserve University to examine the psychosocial experiences of teenagers who have been seen by our craniofacial teams. We are requesting your participation in this study because you are a teen with a craniofacial condition, and was seen by our craniofacial team.

Research Statement

We are currently conducting a study entitled ***"Social-emotional Coping in Adolescents with Craniofacial Conditions"*** that compares the psycho-social adjustment of adolescents with craniofacial conditions to that of adolescents without craniofacial conditions. The following questionnaire is collected for the purpose of research.
The following questionnaire is collected for the purpose of research. The study has 3 components, (3 questionnaires), one to be filled by your parent, one by you (without assistance and in a private place), and one by one of your teachers that you and your parent may recommend.

The questionnaire takes around 15 minutes to complete. It includes questions about social and behavioral functions. Also, we would request from you and your parents to recommend one teacher that you think can fill the same questionnaire on your behalf.

If you are comfortable with us contacting one of your teachers, we would like to have your permission to do so. We will contact the teacher, and explain to them the purpose of our study, and have them fill out the teacher version of the questionnaire. We will collect our data on a secure web portal, to protect the privacy of your information. However, the teacher will preserve the right to deny participation. In that case, your enrollment will not be affected.
If you not wish for us to contact one of your teachers, you can still participate in the study.

Risks and discomforts

There is no risk of physical harm to being in this study, but there is a small risk of loss of privacy and confidentiality which we are protecting against by careful handling of all information. We will use secure web portals for the data collection and storage to protect the privacy of our information.

Benefits

Although there are no direct benefits to our participants, we are anticipating a significant benefit to the community, especially those affected with craniofacial conditions or cleft lip and palate. Our findings would allow us to recommend better support systems for schools and parents and help them implement prevention initiatives, with the long-range goal of developing evidence-based intervention strategies by school personnel and pediatric psychologists.

Confidentiality

Efforts will be made to keep the personal information in your research record private and confidential, but absolute confidentiality cannot be guaranteed. Your name will be removed from all documents and replaced with a code. The University Hospitals Cleveland Medical Center Institutional Review Board may review your study records. The UH REDCap will be used for data collection and storage. It is a secure web application for building and managing online surveys and databases that serves to protect the confidentiality and privacy of your information.

Compensation

You will receive a $10 Amazon gift card for participation, upon completion of the questionnaires.

Contact Person

If you have any questions regarding the study, concerns or complaints about the study in the future, please contact the primary investigator, Dr. Rany Bous. E-mail: rmb184@case.edu. Cell: 440-832-9405.

If the researchers cannot be reached, or if you would like to talk to someone other than the researcher(s) about; concerns regarding the study; research participant's rights; research-related injury; or other human subject issues, please call the University Hospitals Cleveland Medical Center's Research Subject Rights phone line at (216) 983-4979 or write to: The Associate Chief Scientific Officer, The Clinical Research Center, University Hospitals Cleveland Medical Center, 11100 Euclid Avenue, Lakeside 1400, Cleveland, Ohio, 44106-7061.

Voluntary Participation and Right to Withdraw

Participation is voluntary, refusal to participate will involve no penalty or loss of benefits to which the participant is otherwise entitled and you may discontinue participation at any time. Please note that there will be no additional obligation after filling out the survey and that no personal information will be collected to tie you to your participation in the study. You can withdraw at anytime.

To ensure maximum confidentiality, we have replaced your name with the following code: "…". Please use this code to fill the form.

If you have any questions about the study, please contact Rany Bous, BDS at 440-832-9405 or rmb184@case.edu.
Thank you for taking the time to read this, your participation is highly appreciated.

Sincerely,

Rany Bous
Orthodontic department – School of Dental Medicine
Case Western Reserve University
440-832-9405
rmb184@case.edu

23. Teacher recruitment letter

Dear teacher/ Mr. "‥"/ Ms. "…",

We are conducting a study at the University Hospitals Craniofacial Team/Case Western Reserve University to examine the psychosocial experiences of teenagers who have been seen by our craniofacial teams and orthodontic clinics. The study is entitled ***"Social-emotional Coping in Adolescents with Craniofacial Conditions"*** that compares the socio-emotional adjustment of adolescents with craniofacial conditions, to that of adolescents without craniofacial conditions. The following questionnaire is collected for the purpose of scientific research. You are being asked to participate in this study because you are a teacher of a teen with a craniofacial condition or a teacher of a teen without a craniofacial condition seeking orthodontic treatment at our clinic.

One of your students, "name", is one of our participants and we got permission and recommendation from your student and his/her parents to enroll you in our study to get a teacher's perspective.

The main purpose of this study is to identify and compare the difficulties and strengths that face teens with craniofacial conditions compared to non-affected ones. Our secondary objectives are to identify support systems that might help them cope better. Teachers offer an adult's view of the child in relation to a relatively large peer group of the same age. Also, social encounters outside home can be more difficult for the affected patients, and thus we believe that teacher reports can be helpful in identifying the behavioral functioning of adolescents.

The questionnaire takes around 15 minutes to complete. It includes questions about social and behavioral functioning of adolescents. We are only contacting you after obtaining the permission of your student and their parent, and they recommended you as the most appropriate teacher to participate. If you accept to be a part of our research, we will ask you to fill the questionnaire.

The study has 3 components, (3 questionnaires), one to be filled by the parent, one by the child (without assistance and in a private place), and one by yourself, as the teacher chosen by the parent to participate.

Our purpose is to utilize the data to better understand the psycho-social strengths and difficulties that face teenagers, recommend better support systems for schools and parents and help them implement prevention initiatives. That comes with the long-range goal of developing evidence-based intervention strategies by school personnel and pediatric psychologists.

Please note that there will be no additional obligation after filling out the survey. You can withdraw at any time or deny participation. If you deny participation, your student may still participate in our study. Your response is completely private and will not be revealed to the parents or your student.

Risks and discomforts

There is no risk of physical harm to being in this study, but there is a small risk of loss of privacy and confidentiality which we are protecting against by careful handling of all information. We will use secure web portals for the data collection and storage to protect the privacy of our information.

Benefits

Although there are no direct benefits to our participants, we are anticipating a significant benefit to the community, especially those affected with craniofacial conditions or cleft lip and palate. Our findings would allow us to recommend better support systems for schools and parents and help them implement prevention initiatives, with the long-range goal of developing evidence-based intervention strategies by school personnel and pediatric psychologists.

Confidentiality

Efforts will be made to keep the personal information in your research record private and confidential, but absolute confidentiality cannot be guaranteed. The University Hospitals Cleveland Medical Center Institutional Review Board may review your study records. The UH REDCap will be used for data collection and storage. It is a secure web application for building and managing online surveys and databases that serves to protect the confidentiality and privacy of your information. Your name and contact info will be deleted from our records once we receive your questionnaire.

Compensation

Each participating teenager will get a $10 Amazon gift card each, as a token of appreciation for being a part of our research.

Voluntary Participation and Right to Withdraw

Participation is voluntary, refusal to participate will involve no penalty or loss of benefits to which the participant is otherwise entitled and the participant may discontinue participation at any time.

We would be grateful if you would be willing to participate in this survey, as this will promote our understanding of psycho-social functioning among teenagers. If you consent to this project, please follow the link below to answer the questionnaire on a secure portal. If you are unsure how to answer any of the questions, it is acceptable to leave it blank.

To ensure maximum confidentiality, we have replaced your student's name with the following code: "....". Please use this code to fill the form.

Please go to the following link to proceed to the teacher questionnaire:

If you have any questions about the study, please contact Rany Bous, BDS at 440-832-9405 or rmb184@case.edu.
Thank you for taking the time to read this, your participation is highly appreciated.

Sincerely,

Rany Bous
Orthodontic department – School of Dental Medicine
Case Western Reserve University
440-832-9405
rmb184@case.edu

24. **Table 8.** Comparison between our CON and US normative data

	CON (n=55)	US Norms (n=9878)	Sig.*
Total Difficulties	6.3 ± 4.6	7.1±5.7	NS
Emotional problems	1.8 ± 2.0	1.6±1.8	NS
Conduct problems	0.9 ± 1.2	1.3±1.6	NS
Hyperactivity	2.3 ± 2.4	2.8±2.5	NS
Peer Problems	1.3 ± 1.1	1.4±1.5	NS
Prosocial	8.3 ± 1.7	8.6±1.8	NS
Impact on QOL	0.6 ± 1.5	0.4±1.3	NS

* = Independent T-test significance at a p-value of <0.05
NS= No significant differences

Showing an appropriate control group, that functions similar to the normal population. This also implies that orthodontic patients function is in line with normative data