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Dental anomalies associated with cleft lip and palate in Northern Finland

ABSTRACT

Aim *Despite the reported occurrence of dental anomalies of cleft lip and palate, little is known about their prevalence in children from Northern Finland with cleft lip and palate. The aim was to investigate the prevalence of dental anomalies among patients with different types of clefts in Northern Finland.*

Materials and methods *Design and Statistics: patient records of 139 subjects aged three years and older (with clefts treated in Oulu University Hospital, Finland during the period 1996-2010 (total n=183) were analysed for dental anomalies including the number of teeth, morphological and developmental anomalies and their association with the cleft type. The analyses were carried out using Chi-square test and Fisher's exact test. Differences between the groups were considered statistically significant at p values < 0.05.*

Results *More than half of the patients had clefts of the hard palate, 18% of the lip and palate, and 13% of the lip. At least one dental anomaly was detected in 47% of the study population. Almost one in three (26.6%) subjects had at least one anomaly and 17.9% had two or three anomalies. The most common type of anomaly in permanent teeth were missing teeth followed by supernumerary teeth. Supernumerary teeth were significantly more apparent when the lip was involved in the cleft compared with palatal clefts. Missing teeth were less prevalent among those 5 years or younger. The prevalence of different anomalies was significantly associated with the cleft type in both age groups.*

Conclusions *Dental anomalies are more prevalent among cleft children than in the general population in Finland. The most prevalent anomalies associated with cleft were missing and supernumerary teeth.*

Keywords *Dental, Cleft; Dental anomaly; Cleft lip; Cleft palate; Missing teeth; Soft palate cleft; Submucous cleft; Supernumerary teeth.*

Introduction

The most common craniofacial birth defects are clefts of the upper lip and palate [Leite and Koifman, 2009]. In Finland about 130 children are born every year with a cleft from a total of 60,000 newborns for an incidence of approximately 2:1000 [NIHW statistics, 2012]. In Europe the incidence of clefts is 1:1000 [Dixon et al., 2011]. The incidence of clefts in Asia and America is reported to be approximately one in 500 newborns, which is similar to the incidence of isolated palatal clefts reported in Northern Finland [Lithovius et al., 2013 in press]. The frequency of lip and palate clefts is influenced by genetic as well as environmental factors during gestation [Goldschmidt et al., 2010], but aetiology cannot always be tracked. Clefts are common features in some syndromes. Severe clefts may be diagnosed during prenatal ultrasound inspection whereas small clefts are usually diagnosed at birth.

Lip and palate clefts are divided into four main categories: palatal clefts, lip and palatal clefts, lip clefts and lip and alveolar clefts. According to the definition, palatal clefts do not extend to the maxillary alveolus. Submucosal clefts, when the cleft is covered by mucosa, are also classified as a type of palatal cleft. Palatal clefts may involve both the soft and hard palates or only the soft palate. Lip and palatal clefts may extend bilaterally or unilaterally through the alveolar ridge and the lip to the hard and soft palates (Fig. 1).

Isolated palatal clefts are the most common of the cleft types in Northern Finland [Lithovius et al., 2013 in press] comprising over 60% of the clefts needing surgical treatment [Rautio et al., 2010]. The second most common

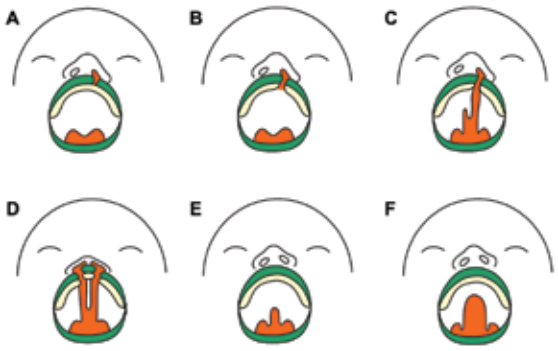

FIG. 1 Different cleft types.
A: Cleft lip. B: Cleft lip and alveolus. C: Cleft lip and palate(unilateral). D: Cleft lip and palate (bilateral). E: Soft Cleft palate. F: Cleft palate (hard and soft).

cleft type involves the lip and palate. Lip clefts, which usually affect the left side, are found in about 15% of the clefts. Bilateral lip clefts are rare. There is evidence of an association of the type of the cleft and dental anomalies especially in recent reports from Asia [Al Jamal et al., 2010; Eslami et al., 2013] but such data is lacking in Northern Finland.

In Finland, the management of cleft patients starts early in the maternity hospital. Surgical treatment is considered when the child is 1 to 3 months old. The core of the cleft treatment is multidisciplinary, being provided by members of the cleft team including oral and maxillofacial, craniofacial and plastic surgery, otolaryngology, phoniatry, speech therapy, paediatric dentistry, orthodontics as well as psychiatry, nursing and genetics. In Finland patients with lip and palate clefts are treated at two centres, Oulu and Helsinki University Hospitals. In Oulu the patients are monitored in the University Hospital regularly until they are 18 years of age. Orthodontic and basic treatment can also be performed at health centers or in the private sector with financial support from the community being free of charges to the individual until the age of 18.

The prevalence of dental anomalies was recently reported to be 11.7% in an article on panoramic radiographs in children of general population aged 5 to 12 years. The most prevalent anomalies were missing and supernumerary teeth, occurring at a rate of 4.63% and 3.31%, respectively [Souchois et al., 2013]. Anomalies in the deciduous dentition are clearly more common in children with clefts than in non-cleft children. Minor morphological disturbances in deciduous teeth are very frequent in children with cleft lip and palate [Pöyry and Ranta, 1985a, b]. The hypothesis was that the prevalence of dental anomalies among cleft patients is higher than in the general population with missing and supernumerary teeth being the most prevalent dental anomalies in permanent dentition.

The aim of this study was to investigate the prevalence of dental anomalies in patients with clefts specifically in Northern Finland considering the age and gender of the subject as well as the cleft type.

Material and methods

Material for this retrospective population based follow-up study was collected from the Oulu University Hospital patient registry, comprehensively covering the period of 15 years (1996-2010) when the cleft patients have been treated in Oulu. The data on patients who had received treatment in Helsinki were not available. Data of those treated in Oulu (n=183) were collected and data of those who were 3 years or older were then analysed together with the other authors.

Patients under 3 years of age were excluded because dental anomalies are not reliably detectable in such a young age group. The data comprised medical histories of the total of 139 cleft patients who fit the above inclusion criteria. The following factors were recorded: date of birth, gender, cleft type, and dental anomalies in permanent teeth. To assess differences in prevalence of anomalies in different age groups, the data were dichotomised: cleft patients older than 5 years and 5 years or younger. This strategy was adopted since children under 3 years of age rarely had radiographs available and some deciduous tooth anomalies might not be clinically visible. Since the majority of the dental anomalies in the study population were anticipated to be in the cleft adjacent areas, and since most cleft adjacent teeth are at least radiographically visible by 5 years of age, the authors accepted this dichotomised approach. The clefts were divided into 7 groups: hard palate cleft, soft palate cleft, right side lip and palate cleft, left side lip and palate cleft, bilateral lip and palate cleft, lip and alveolus cleft, and submucosal cleft. The prevalence of developmental disorders in permanent teeth was reported tooth-wise on the basis of the registered diagnosis code or ICD 10 (K00). Anomalies were reported in the patient files according to clinical or radiographic findings on all available panoramic radiographs, cone beam CT scans, occlusal films or periapical x-rays. Anomalies were further categorised as follows: mineralisation defects (Fig. 2A), morphological changes (Fig. 2B), supernumerary teeth (Fig. 2D), missing teeth (Fig. 2C), gemination and hypoplastic teeth. The anomalies had been registered using Yes/No answers.

Ethical approval

The study was carried out using the Oulu University Hospital patient registry data (permission number 10/2012). The data were analysed without personal identifying details, so no separate permission from the Ethics Committee was required. Permission from the registry holder (Oulu University Hospital) was considered to be sufficient for this retrospective register-based study.

Statistics

The frequencies of different types of clefts and anomalies were analysed from the data according to gender. In analysing the association between the cleft type

FIG. 2A Mineralisation defect.

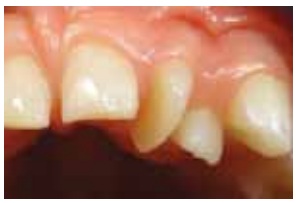

FIG. 2B Morphologic defect at sites 21 and 22.

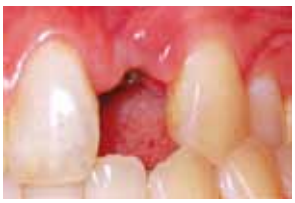

FIG. 2C Missing tooth at site 22 replaced by an implant.

FIG. 2D Supernumerary tooth located between teeth 61 and 62.

Type of the cleft	Male n (%)	Female n (%)	p	Total n (%)
Lip & alveolus	5 (7.6)	4 (5.5)	n.s.	9 (6.5)
Hard palate	29 (43.9)	46 (63.0)	0.024	75 (54.0)
Lip & palate (left)	4 (6.1)	7 (9.6)	n.s.	11 (7.9)
Lip & palate (right)	2 (3.0)	2 (2.7)	n.s.	4 (2.9)
Lip & palate (bilateral)	9 (13.6)	1 (1.4)	0.007	10 (7.2)
Lip	5 (7.6)	4 (5.5)	n.s.	9 (6.5)
Submucous	7 (10.6)	3 (4.1)	n.s	10 (7.2)
Soft palate	5 (7.6)	6 (8.2)	n.s.	11 (7.9)
Total	66 (47.5)	73 (52.5)		139 (100.0)

TABLE 1 Prevalence of the clefts between the genders.

and dental anomaly, the age of the participants was taken into consideration. The association between clefts and developmental dental disorders was evaluated using Chi-squared test or Fisher's exact test. Difference between the groups was considered statistically significant at p values $p<0.05$. Statistical analyses were performed using the program SPSS version 20.0 (SPSS, Inc., Chicago, Il, USA).

Results

The mean age of the study population was 8.2 years (95% Confidence Interval lower boundary 7.68 and upper boundary 8.70; min 3.05, max 15.66). There were 108 patients who were older than 5 years and 31 patients who were 5 years or younger. The study population was slightly dominated by females (52.5%) (Table 1). The most prevalent clefts were those involving the hard palate followed by lip and palate clefts (combining left, right and bilateral), and lip clefts with or without involvement of alveolus. The least prevalent clefts were sub mucous and soft palate clefts. Hard palate clefts were statistically significantly more prevalent among girls than boys, while the opposite was true for bilateral lip and palate clefts (Table 1). The majority of the children were healthy, 18 (12.9%) had a diagnosed syndrome. Those syndromes represented in the study population included Pierre Robin, Treacher Collins, Fragile X, Blepharo-cheilo-dontic syndrome, Kabuki, Apert, CATCH 22, and Down syndromes. Of those with syndromes 66.7% had a hard palate cleft, 16.7% had a sub mucous cleft, 11.2% had a cleft lip and palate, 0% had soft palate cleft and 5.6% had lip cleft. Distribution of different types of clefts was similar among healthy patients and those with syndromes.

At least one dental anomaly was detected in 47% of the 139 cleft patients. Almost one in three (26.6%) had at least one anomaly and 17.9% had two or three anomalies. Missing teeth were the most common dental anomaly followed by supernumerary teeth among cleft patients. Patients with all different cleft types except lip clefts had missing teeth, whereas supernumerary teeth were associated with other cleft types except hard palate and submucous clefts. Cleft patients also commonly had morphological changes and hypoplastic teeth (Table 2). Most anomalies occurred in children with cleft lip and palate, followed by children with hard palate and lip clefts (with or without alveolar cleft). Children with cleft lip and palate had significantly more frequently missing and supernumerary teeth, mineralisation defects and morphological changes as well as hypoplasias than the rest. Children with hard palate cleft had missing teeth, morphological and hypoplastic disorder. Although supernumerary teeth were frequent especially among children with cleft lip in the data, they were not found in any patients with hard palate clefts.

Cleft Type	Missing teeth n (%)	Supernumerary teeth n (%)	Gemination n (%)	Mineralisation defects n (%)	Morphological changes n (%)**	Hypoplastic n (%)*	Total n (%)
Lip and alveolus	2 (5.6)	4 (18.2)*	1 (100.0)	1 (10.0)	3 (15.8)	1 (7.1)	12 (11.8)
Hard palate	17 (47.2)	0 (0.0)***	0 (0.0)	4 (40.0)	6 (31.6)*	4 (28.6)*	31 (30.4)
Lip & palate (left)	7 (19.4)**	6 (27.3)***	0 (0.0)	3 (30.0)*	2 (10.5)	4 (28.6)*	22 (21.6)
Lip & palate (right)	2 (5.4)	1 (4.5)	0 (0.0)	0 (0.0)	1 (5.3)	1 (7.1)	5 (4.9)
Lip & palate (bilateral)	4 (11.1)	4 (18.2)	0 (0.0)	1 (10.0)	4 (21.1)*	1 (7.1)	14 (13.7)
Lip	0 (0.0)	6 (27.3)**	0 (0.0)	0 (0.0)	3 (15.8)	2 (14.3)	11 (10.8)
Submucous	3 (8.3)	1 (4.5)	0 (0.0)	1 (10.0)	0 (0.0)	1 (7.1)	6 (5.9)
Soft palate	1 (2.8)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (1.0)
Total n (%)	36 (35.3)	22 (21.6)	1 (1.0)	10 (9.8)	19 (18.6)	14 (13.7)	102 (100.0)
* p<0.05, ** p<0.01, *** p<0.001							

TABLE 2 Association of the cleft types and prevalence of dental anomalies (139 patients); statistical significance in prevalence between the cleft types.

Children with submucous and especially soft palate clefts had least dental anomalies. Prevalence of dental anomalies was statistically significantly associated with the cleft type (Table 2). There were 86 anomalies among 108 children in the older group and 17 among 31 in the younger group. There were some differences in the prevalence of anomalies between the age groups. The proportions of missing and supernumerary teeth differed the most. Proportion of the missing teeth dominated among the older group whereas that of supernumerary and hypoplastic teeth among the younger ones. The prevalence of supernumerary teeth was significantly associated with the cleft type in both age groups, especially in those involving the lip (Table 3).

Discussion

The results indicate that developmental dental disorders are more common among cleft patients than healthy individuals. For instance, in the general population missing teeth are found in 6 to 9% [Avellán et al., 2010] whereas in this study 25% of the cleft patients had missing teeth. In cleft patients older than 5 years the respective frequency was even higher (>30%). Similarly supernumerary teeth were found in 0.4% of Finnish children aged 3-4 years [Järvinen and Lehtinen, 1981], whereas among cleft patients in this study the prevalence was 15.8%; being the same also in patients older than 5. In comparison the prevalence of dental anomalies

Patients older than 5years (n=108)							
Cleft Type	**Missing teeth n (%)**	**Supernumerary teeth n (%)**	**Gemination n (%)**	**Mineralisation defects n (%)**	**Morphological changes n (%)****	**Hypoplastic n (%)***	**Total n (%)**
Lip and alveolus	1 (3.0)	3 (17.6)*	1 (100.0)	1 (12.5)	2 (11.8)	1 (10.0)	14 (16.3)
Hard palate	17 (51.5)	0 (0.0)***	0 (0.0)	3 (37.5)	6 (35.3)	3(30.0)	29 (33.7)
Lip & palate (left)	7 (21.2)**	5 (29.4)**	0 (0.0)	3 (37.5)*	2 (11.8)	4 (40.0)**	21 (24.4)
Lip & palate (right)	2 (6.1)	1 (5.9)	0 (0.0)	0 (0.0)	1 (5.9)	1 (10.0)	5 (5.8)
Lip & palate (bilateral)	4 (12.1)	4 (23.5)*	0 (0.0)	1 (12.5)	4 (23.5)*	1 (10.0)	14 (16.3)
Lip	0 (0.0)	3 (17.6)*	0 (0.0)	0 (0.0)	2 (11.8)	0 (0.0)	3 (3.5)
Submucous	2 (6.1)	1 (5.9)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	
Soft palate	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0
Total n (%)	33 (38.4)	17 (19.8)	1 (1.2)	8 (9.3)	17 (19.8)	10 (11.6)	86 (100.0)
* p<0.05, ** p<0.01, *** p<0.001							
Patients 5 years or younger or 3-5 years (n=31)							
Cleft Type	**Missing teeth n (%)**	**Supernumerary teeth n (%)**	**Gemination n (%)**	**Mineralisation defects n (%)**	**Morphological changes n (%)****	**Hypoplastic n (%)***	**Total n (%)**
Lip and alveolus	1 (33.3)	1 (20.0)*	0 (0.0)	0 (0.0)	1(50.0)	0 (0.0)	3 (17.6)
Hard palate	0 (0.0)	0 (0.0)*	0 (0.0)	1 (50.0)	0 (0.0)	1 (25.0)	2 (11.8)
Lip & palate (left)	0 (0.0)	1 (20.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (20.0)	2 (11.8)
Lip & palate (right)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Lip & palate (bilateral)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Lip	0 (0.0)	3 (60.0)**	0 (0.0)	0 (0.0)	1 (50.0)	2 (50.0)	6 (35.3)
Submucous	1 (33.3)	0 (0.0)	0 (0.0)	1 (50.0)	0 (0.0)	1(25.0)	3 (17.6)
Soft palate	1 (33.3)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (5.9
Total n (%)	3 (17.6)	5 (29.4)	0 (0.0)	2 (11.8)	2 (11.8)	5 (29.4)	17 (100.0)
* p<0.05, ** p<0.01, *** p<0.001							

TABLE 3 Association of the cleft types and dental anomalies in two different age groups (5 years and younger and older than 5 years).

among Turkish cleft patients was 96.7%. Almost all of these patients were reported to have at least one dental anomaly [Akcam et al., 2010]. In this study about half of the cleft patients from Northern Finland were reported to have at least one dental anomaly.

Based on this study, it may be concluded that the prevalence of dental anomalies is strongly influenced by the type of the cleft. Supernumerary teeth are found in all other types of clefts except those involving only the palate. They also seem to manifest earlier than missing teeth. Missing teeth can clearly be found significantly more often in patients with a unilateral or bilateral lip and palate clefts than in patients with soft palate or sub mucous clefts. In uni- or bilateral lip and palate clefts the cleft cuts through the alveolar bone and the development of the teeth is often affected, causing agenesis or deformation of teeth [Eslami et al., 2013].

The prevalence of dental anomalies of the cleft patients seems to vary geographically. As to prevalence of supernumerary teeth among the cleft patients, the results of this study (15.8%) are in agreement with those of Al Jamal et al. [2010], who reported supernumerary teeth among 16.7% of the cleft patients in Jordan. However, the prevalence of missing teeth reported by Al Jamal et al. [2010] was much higher (66.7%) than that found in the present study. This was also true among Chinese cleft patients (12-16 years) with prevalence of 57.6% of missing teeth [Wong et al., 2012]. Missing teeth were reported to be the most frequent dental anomaly also among Iranian and Turkish cleft patients (70.8–97.1%) [Akcam et al., 2010; Eslami et al., 2013]. The authors could not find any Northern European studies on prevalence of dental anomalies among cleft patients. This enhances the value of the present study.

Morphological changes as well as hypoplastic teeth comprised about one third of the anomalies. Satisfactory appearance is an issue of major concern for development of good self-esteem. Therefore these anomalies and especially anterior morphological changes should be treated as early in life as possible. If permanent treatment cannot be carried out, temporary solutions should be considered. Composites combined with fiber offer a good temporary treatment alternative even for developing dentition.

In this study group the clefts are more common among girls than boys. The girls have significantly more frequently hard palate clefts whereas the boys have significantly more frequently more serious bilateral lip and palate clefts. Lip and palate clefts are one symptom among many others which are present in more than 300 syndromes [Rautio et al., 2010; Cameron and Widmer, 2008]. In the current study the proportion of syndromic children with clefts was 11.5%, being similar to that reported on Ireland (14.8%) [Mc Donnell et al., 2013]. According to our results the clefts among the children with syndromes are not more complex than in non-syndromic children and the prevalence of dental anomalies is similar in both groups as well. It seems that genetics play an important role in the aetiology of the clefts. However, many other congenital deformities have been found to be linked with environmental factors such as maternal smoking and alcohol use during pregnancy [Alzenbud et al., 2013]. Studies on cleft lip and palate patients indicate that clefts may also be associated with diet, vitamin consumption and drug use during pregnancy. Low birth weight is also connected with the incidence of palate clefts [Gonzales et al., 2008]. In a cross-sectional study conducted in Pakistan, the aetiological factors of the clefts could not be tracked in 82% of the cases. In 6% of the cases the cause was environmental and in 12% it was genetic. In that study 18% of the patients also had dental anomalies. Environmental factors vary in different parts of the world. For instance the prevalence of diabetes, a considerable risk factor during pregnancy for development of clefts [Yaqoob et al., 2013], varies greatly. The presence of dental anomalies, such as molar incisor hypomineralisation disorder (MIH) has been connected with environmental toxins. The original aetiological factor for both the cleft and dental anomalies can be the same, but the physical existence of the cleft may also affect the developing tooth buds causing a dental anomaly. This might explain the wider prevalence of dental anomalies among severe clefts and their non-existence among the less severe soft palate and sub mucous clefts. The youngest children operated in the Oulu University Hospital were excluded from the study group being aware of dental anomalies becoming manifest only later on in life. To assess differences in the prevalence of anomalies in different age groups, the authors decided that the data were to be dichotomised, having cleft patients older than 5 years and 5 years or younger as the cut-off point. The majority of anomalies were anticipated to be in cleft-adjacent teeth. The crowns of the cleft-adjacent teeth being incisors and canines, they are generally developed to the point of being radiographically detectable by the age of 5 years. The authors understood the negative side of the timing of this dichotomous division, being that some findings in the non-cleft adjacent premolar areas of the jaws might not be detected. This meant that the true incidence of dental anomalies might in fact be underestimated by this analysis. Further sub stratifying the data to more age groups did not add to the analysis. Generally studies on dental anomalies are cross-sectional on a variety of older age groups. There are not many studies that compare these younger and older age groups regarding dental anomalies among cleft patients [Pöyry and Ranta, 1986a, b; Pöyry et al., 1989]. There was a significant difference in prevalence of missing teeth between the two age groups. Teeth were detected as missing less frequently among those 5 years or younger than in the older group. The disorders of the permanent dentition become manifest only after the permanent dentition develops, for example, all teeth excluding wisdom teeth develop generally by the age 13. This explains the lower frequency of missing

teeth in the younger age group in the present study. Also mineralisation defects and morphological changes may be diagnosed reliably only at a later age. However, detection of any anomalies as early as possible is beneficial to estimate the patient's future dental treatment plan needs. Documenting the presence of dental anomalies is important in predicting the costs related to dental care used for understanding the budgetary projections of such comprehensive programmes.

The treatment of the clefts is defined by its type. Isolated lip clefts and separate palate clefts often do not require any treatment other than primary surgery. Lip and palate clefts usually require several operations, such as a bone graft and possibly orthognathic surgery to correct jaw growth deficits. Severe clefts can also affect tooth development and the development of speech and treatment of children with clefts usually includes speech therapy possibly combined with speech improving operations. Regarding the dentition Pöyry et al. [1989] have shown already in the 1980s that a mean delay of tooth development of the cleft patients (3-9 years) with or without a cleft palate is 6 months, which decreases to two months in the age of group of 8-14 years. Involvement of the palate makes the delay longer compared with the cleft lip only [Pöyry and Ranta, 1986a; Pöyry et al., 1989]. The early timing of tooth development is close to that reported for healthy children [Pöyry and Ranta, 1986b]. Many cleft patients also have problems with their ears, mostly with recurrent acute otitis media. Therefore, in many cases, cleft patients also need myringotomy surgery. Cleft patients also require orthodontic treatment in addition to treatment for morphological anomalies [Rautio et al., 2010]. The edentulous space left by the cleft and missing teeth can be restored prosthodontically after growth has ceased or with orthodontic treatment during growth. Also surgical treatment, such as tooth transplantation, can be used for filling gaps with other teeth. Dental implants can be used later after growth cessation during adulthood [Sedlackova et al., 2011; Aizenbud et al., 2013]. Clefts require long and challenging multidisciplinary treatment plans and their precise execution. The treatments are physically and mentally demanding both for the patients and their parents as well as the treatment team.

Conclusion

The prevalence of dental anomalies among cleft patients is higher than in the general population and missing and supernumerary teeth are the most prevalent dental anomalies in cleft patients in the Northern Finnish population. Clefts are significantly associated with gender as girls dominate. The most common cleft type are hard palate clefts, which fortunately are less serious and require fewer surgeries than clefts involving the alveolar ridge. The lip and palate clefts are more common on the left side in this population; the cause for this predilection being unknown. Children with the most severe clefts have the greatest number of dental anomalies. Clefts are often associated with developmental disorders of teeth presenting as oligodontia, supernumerary teeth, hypoplasia or hypomineralisation. These observations should be taken into consideration both in treatment planning and in making budgetary projections.

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