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ORIGINAL ARTICLE

Pattern of clefts and dental anomalies in six-year-old children: a retrospective observational study in western Norway

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ABSTRACT

Objectives: Clefts of the lip and/or palate (CL/P) are the most common congenital disorders of the head and neck. In Norway, the incidence is 1.9/1000 live births. The aim of this study was to investigate the frequency and distribution of various types of clefts and dental anomalies in patients treated by the cleft lip and palate (CLP) team in Bergen, Norway.

Material and methods: The material comprised the records of patients 6 years of age, examined by the CLP team in Bergen from spring 1993 to autumn 2012, incomplete records were excluded. The records of 989 patients were analysed, using frequencies and Chi-square test to compare differences in percentages between groups.

Results: The gender distribution was 58.8% male and 41.2% female. Isolated cleft palate (CP) was the most common condition (39.5%). Clefts of the lip, jaw and palate (CLP) constituted (30%) of cases and (30.5%) had isolated cleft lip (CL). The frequencies of agenesis, supernumerary and peg-shaped teeth were (36.5%), (17.8%) and (7.5%), respectively. Over 50% of the study population were diagnosed with one or more malocclusion. Of the CLP patients, 61.4% had Angle Class III occlusion. Statistical analysis disclosed a positive association of agenesis with Class III occlusion (OR $=1.8$, $p \leq 0.001$).

Conclusions: The findings supported the hypothesis that the distribution of dental anomalies and occlusal disorders varied among patients with CL, CP and CLP. In patients with cleft, there is a twofold chance to get Class III malocclusion in the presence of agenesis.

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Introduction

Clefts of the lip and/or palate (CL/P) are the most common congenital disorders of the head and neck. The cleft results from incomplete fusion between the lip and primary palate with the secondary palate. The cleft of the lip can be either unilateral or bilateral.[1] In Norway, the incidence is 1.9 per 1000 live births.[2]

Reports of CL/P prevalence vary. In a cohort from East Ireland the total birth prevalence of all orofacial clefts was 16.0 per 10,000 births.[3] Souza and Raskin reported a prevalence of 1/1010 live births with 21% cleft palate (CP), 24% cleft lip (CL) and 55% with cleft lip and palate (CLP).[4] A Finish study reported that CP was more frequent in females (63.3%) and CLP was more frequent in males (62.5%). In the same study, a family history of clefts was found in 20.1% of patients.[5] In 2003, WHO reported that in 1.8 million Norwegian live births for the period between 1967 to 1998, the incidence of facial clefts was 1.9/1000.[2] There is a lack of recent published data on the incidence of CL/P in Norway. Previously, it was reported that individuals born with clefts have more dental anomalies in the permanent dentition than unaffected individuals.[6–8]

Occlusal anomalies have been reported in patients with CL/P.[9] In a previous study, normal occlusion was observed in 50% of the control group and in 30% of the cases with cleft.[9] In a study among Brazilian children between 6 and 12 years of age, the clefts were classified as pre- and trans-incisal foramen cleft. Angle Class III was found among pre- (16.7%) and trans- (21.6%) incisal foramen cleft patients. Angle Class II was present in pre- and trans-incisal foramen cleft patients in percentage of 50% and 70%, respectively.[10] Baek and coauthors stated that subjects with CP and CLP were more likely to have a Class III occlusion with a negative overjet than those with CL only.[11]

Hypodontia has been reported in 70% of individuals with complete bilateral CLP: the maxillary lateral incisor was the most affected tooth. The same study reported supernumerary teeth in 11.7% of the subjects.[12] Agenesis of at least one tooth was reported in approximately 60% of patients aged 10.5–13.5 years with complete bilateral CLP. The teeth most frequently missing were the maxillary lateral incisors, the maxillary and mandibular second premolars.[13] There are no recent studies of dental and occlusal abnormalities in Norwegians with CL/P.

Successful treatment of CL/P often presents a clinical challenge, particularly when the condition is complicated by dental and occlusal anomalies. Improved understanding of the relationship between such anomalies and CL/P would

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facilitate appropriate treatment planning and evidenced-information for counselling of cleft patients and their parents. Our hypothesis is that the distribution of dental anomalies and occlusal disorders differ according to the type of cleft.

The aim of the present study was to: study the frequency and distribution of different cleft types among children born in western part of Norway between 1987 and 2006 and to assess cleft types' possible relation with dental anomalies and occlusal disorders in the study subjects.

Material and methods

The study was designed as a retrospective observational cross-sectional study. Data were collected for the period 1993–2012, from records of 6-year-old patients with CL/P attending the dental clinic at the Oral Health Center of Expertise/Western Norway, Hordaland. The CLP patients are treated and followed up by CLP team in Bergen. The follow-up period varies according to the patient' treatment needs. As a routine all patients, regardless of diagnosis have control at the age of 6- and 16-year old. The 6-year-old group was enrolled in the present study to detect occlusal disorders prior to the initiation of orthodontic treatment.

The subjects comprised 1022 CL/P children 6 years of age, born between 1987 and 2006. The inclusion criteria comprise of clinical records of the children with cleft lip/palate who attended the 6-year follow-up clinic. To be included in the study, the record should contain; the date of birth, gender, cleft type and dental anomalies in permanent teeth. The exclusion criteria were incomplete registers due to severe syndromes and or lack of cooperation. Thus, thirty three patient's records were excluded from the study.

Anomalies were reported in the patients' files according to the clinical or radiographic findings on lateral, panoramic, occlusal films and periapical X-rays and dental casts. Data including type of cleft, dental and occlusal abnormalities were retrieved from patient records. In conditions where diagnosis of dental anomalies was not well-defined, the treating orthodontists had the possibility to verify the diagnosis through patient's electronic journal record.

ICD-10 codes were used to record the presence of the different types of cleft. The clefts were divided as following: The clefts that involve the palate are classified into hard palate cleft (Q 35.1), soft palate cleft (Q 35.3), hard and soft palate cleft (Q 35.5) as well as unspecific cleft palate (Q 35.9). The clefts that involve the lip are classified into cleft lip bilateral (Q 36.0), midline cleft lip (Q 36.1) and cleft lip unilateral (Q 36.9). The hard palate cleft in combination with bilateral (Q 37.0) or unilateral (Q 37.1) cleft lip. Soft palate cleft in combination with bilateral (Q 37.2) or unilateral (Q 37.3) cleft lip. Soft and hard palate cleft in combination with bilateral (Q 37.4) or unilateral (Q 37.5) cleft lip. The cleft types were also classified into CL, CP and CLP in order to explore potential relations between the type of clefts, and dental abnormalities.

The developmental disorders in permanent teeth were described by using radiographs and clinical records and reported using the registered diagnosis code or ICD 10. Anomalies were categorized as follows: missing teeth, supernumerary teeth, inverted and peg-shaped teeth. Not commonly found defects with undefined morphologic variation were named as rare defects such as taurodontism, microdontia and hypoplastic teeth.

The malocclusion was defined by sagittal, vertical and transversal relation as well as by space conditions. The sagittal relation was described as edge to edge, Class II combined with positive overjet and Class III combined with negative overjet. The Angle classification in combination with the anterioposterior relation in the front segment was used to assess the sagittal relation. The first molar was used as an indicator tooth and since our participants have mixed dentition the deciduous canine was used if the first molar was not erupted. The transversal malocclusion, decided by evaluating the relation between mandible and maxilla in the lateral segment, classified as cross bite either uni- or bi-lateral and scissor bite. The vertical malocclusion was determined by measuring the distance between the incisal edge of the upper and lower front teeth and a distance of (0–5 mm) was considered normal. A higher distance than 5 mm was considered as deep bite, and negative overlapping was an open bite. When the front teeth were missing or not fully erupted, the evaluation was then performed using the curvature of the jaw seen from dental model casts and from lateral radiographs. Identification data, such as patients' names and unique personal identification numbers, were masked for the investigators. Ethical approval was granted by the Norwegian Social Science Data Services (NSD).

Statistical methods

Data management and analyses were performed using the program Statistical Package for the Social Sciences version 20.0 (SPSS, Inc., Chicago, IL). The frequencies and percentages were used at the univariate level to describe the distribution of the different types of clefts. Chi-square test was used in the bivariate analyses to identify possible associations between gender, cleft types, occlusion and dental anomalies. Association between malocclusions and dental anomalies was also assessed by Chi-square test using odds ratios as a measure of likelihood of occurrence. Difference between the groups was considered statistically significant at p values $p < 0.05$ with a confidence interval of 95%.

Results

The results include 989 records of children born in Western Norway diagnosed with CL/P. More boys 58.7% ($n = 581$) were affected than girls 41.3% ($n = 407$). Approximately 40% of the study population had CP and the distribution as follows: Q35.3 ($n = 162$), Q35.5 ($n = 178$) and Q35.9 ($n = 48$). According to the ICD-10 classification the unilateral CL (Q36.9) is the most frequent 27.9% ($n = 276$) diagnose among the cleft subgroups (Figure 1(A)). Analysis of the data (Figure 1(B), disclosed the most common cleft type among girls to be cleft of the soft and hard palate (Q 35.5) followed by unilateral CL (Q 39.6). Among boys, the most frequently observed subgroups were unilateral CL (Q 39.6) and soft and

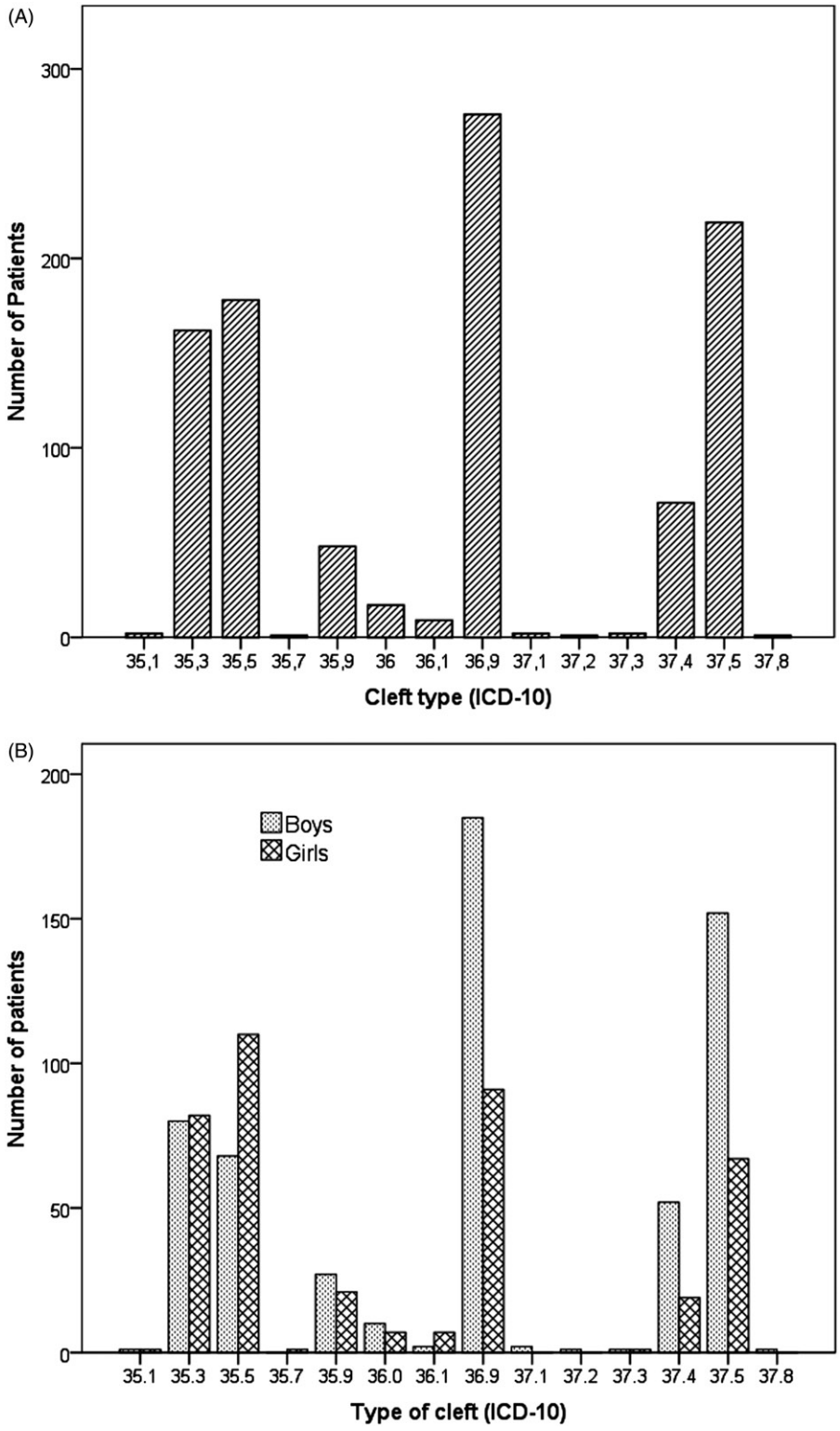

Figure 1. Distribution of cleft types according to the ICD-10 classification (A), gender distribution (B).

hard CP in combination with unilateral CL (37.5). The left side clefts were detected predominantly (Data not shown). Figure 2 illustrates that the yearly distribution of different types of cleft varies throughout the study period.

The most frequent dental anomalies in the study population were hypodontia affecting more females and supernumerary teeth affecting more males. Inverted and peg-shaped teeth affected predominantly males. Undefined defects presented as rare anomalies in 3.9% of the study population (Table 1).

In all patients, 36.5% ($n = 361$) had one or more missing teeth. The highest percentage of hypodontia was for the maxillary second premolars 30% ($n = 297$), followed by the maxillary lateral incisors 21.9% ($n = 217$). In the mandible, hypodontia of the left and right second premolars was the most common: 6.5% and 7.1%, respectively.

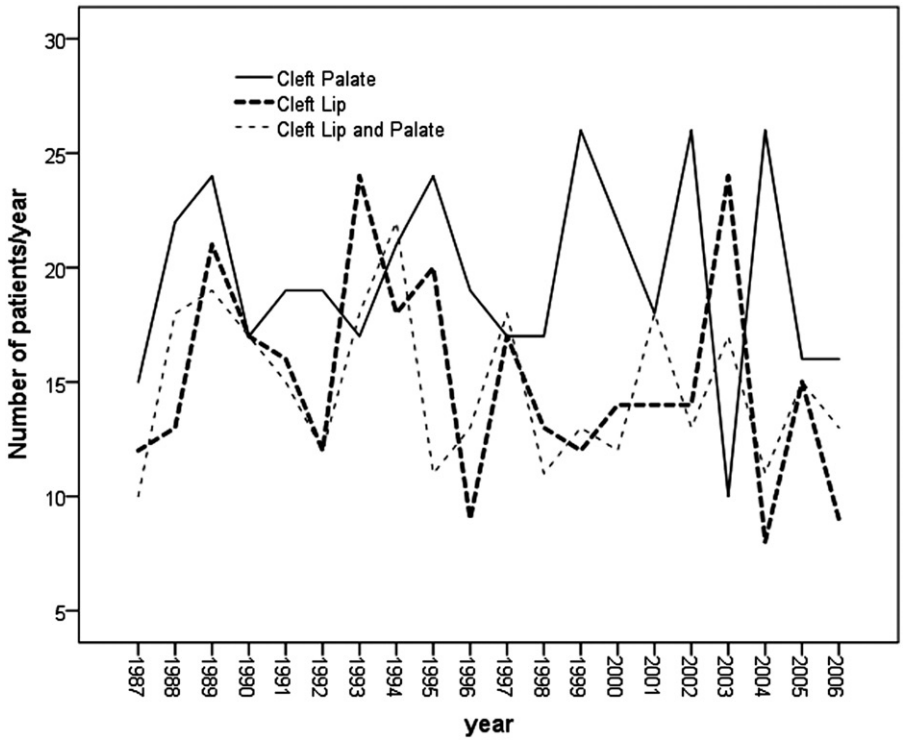

Figure 2. Prevalence of different types of cleft among children born in Western Norway between 1987 and 2006.

Table 1. Frequency and percentage distribution of different CL/P types, dental anomalies and malocclusions by gender.

Gender	Male *n* (%)	Female *n* (%)	Total *n* (%)
CL	197 (33.8)	105 (25.8)	302 (30.5)
CP	176 (30.2)	215 (52.8)	391 (39.5)
CLP	209 (35.9)	87 (21.4)	296 (30.0)
Agenesis	191 (32.9)	170 (41.9)	361 (36.5)
Supernumerary teeth	130 (22.4)	46 (11.3)	176 (17.8)
Inverted teeth	33 (5.7)	14 (3.4)	47 (4.8)
Peg-shaped teeth	50 (9.1)	24 (6.4)	74 (7.5)
Rare anomalies	24 (4.1)	15 (3.7)	39 (3.9)
Malocclusion	298 (51.4)	208 (51.1)	506 (51.2)
Crowding	61 (10.5)	64 (15.7)	125 (12.7)
Angle Class II	22 (3.8)	20 (4.9)	42 (4.3)
Angle Class III	103 (17.7)	55 (13.5)	158 (16)
Edge to edge	43 (7.4)	40 (9.8)	83 (8.4)
Open bite	10 (1.7)	9 (2.2)	19 (1.9)
Deep bite	18 (3.1)	10 (2.5)	28 (2.8)
Bilateral crossbite	43 (7.4)	22 (5.4)	65 (6.6)
Unilateral crossbite	112 (19.3)	45 (11.1)	157 (15.9)
Scissors bite	2 (0.3)	2 (0.5)	4 (0.4)

Table 2. Distribution of dento-occlusal anomalies according to type of cleft. Undefined tooth anomalies were registered as rare anomalies.

	CL *n* (%)	CP *n* (%)	CLP *n* (%)
Agenesis	57 (15.8)	134 (37.1)	170 (47.1)
Supernumerary teeth	128 (72.7)	3 (1.7)	45 (25.6)
Inverted teeth	13 (27.7)	11 (23.4)	23 (48.9)
Peg-shaped teeth	40 (54.1)	9 (12.2)	25 (33.8)
Rare anomalies	16 (41.0)	4 (10.3)	19 (48.7)
Malocclusion	90 (17.8)	206 (40.7)	210 (41.5)

Table 3. Distribution of malocclusions according to cleft type.

	CL *n* (%)	CP *n* (%)	CLP *n* (%)
Crowding	20 (16.0)	81 (64.8)	24 (19.2)
Angle Class II	10 (23.8)	28 (66.7)	4 (9.5)
Angle Class III	19 (12.0)	42 (26.6)	97 (61.4)
Edge to edge	12 (14.5)	33 (39.8)	38 (45.8)
Open bite	4 (21.1)	11 (57.9)	4 (21.1)
Deep bite	11 (39.3)	12 (42.9)	5 (17.9)
Bilateral crossbite	6 (9.2)	10 (15.4)	49 (75.4)
Unilateral crossbite	20 (12.7)	38 (24.2)	99 (63.1)
Scissors bite	2 (50.0)	2 (50.0)	0 (0)

Seventeen percent of the patients with cleft had one or more supernumerary teeth in the region of the maxillary lateral incisors. The maximum number of supernumerary teeth in one patient was four teeth. The highest percentage was found in subjects with CL (72.7%) (Table 2).

The data showed that 7.5% of subjects had one or more peg-shaped teeth ($n = 74$). The most frequently affected teeth were the maxillary left and right lateral incisors (57.5% and 37.9%, respectively). The condition affected 54.1% of subjects with CL and 33.8% of those with CLP (Table 2).

More than half of patients (51.2%) had at least one occlusal anomaly and 7% exhibited three types of malocclusion concurrently (Table 1). Crowding of the teeth was the most frequent (64.8%) occlusal anomaly in CP subjects (Table 3). In regard to sagittal relations, Angle Class III occlusion with a negative overjet and edge to edge occlusion were most frequent among patients with CLP 61.4% and 45.8%, respectively. The results for the vertical relations showed that in the study population 2.8% had deep bite and 1.9% had open bite (Table 1). The transversal deviation including unilateral and bilateral crossbites was common among CLP subjects (Tables 1 and 3); males 19.3% ($n = 112$) were affected more frequently by unilateral crossbite than females 11.1% ($n = 45$). Statistical analysis disclosed a positive association of agenesis with Class III (OR $= 1.8$, $p \leq 0.001$), edge-to-edge malocclusion (OR $= 1.9$, $p \leq 0.01$) and unilateral crossbite

(OR =1.6, $p \leq 0.01$). Further analysis disclosed a negative association of supernumerary tooth anomaly with Class III occlusion (OR =0.6, $p \leq 0.02$), and dental crowding (OR =0.3, $p \leq 0.00$).

Discussion

The study was undertaken in order to investigate the pattern of cleft types, dental anomalies and occlusal disorders in patients treated by the cleft lip and palate (CLP) team in Bergen, Norway. Analysis of gender distribution disclosed that the majority of the cases with CL/P were boys. A similar gender distribution has been reported among Norwegian newborns diagnosed with CL/P.[14,15] In a previous review, including 110 epidemiological reports on CL/P in Europe, it was found that males are more frequently affected than females, 58% versus 42%.[16] In accordance with the existing literature,[17] the present study showed that all types of cleft, with the exception of soft and hard cleft palate (Q35.3 and Q35.5) and cleft of the midline of the upper lip (Q36.1), occurred more frequently in boys than in girls.

In this report, agenesis of one or more teeth was recorded in 36.5% of the subjects. This finding is in accordance with a Dutch study reporting agenesis in 39.7% of the study population.[18] In another study, a higher percentage (77%) have been reported.[19] In the present study, the prevalence of hypodontia in CL patients was lower than in CLP patients. This finding is in accordance with previous reports suggesting that agenesis is more frequently associated with more complicated types of cleft.[20] In agreement with a previous study, the percentage of agenesis was higher in the maxillary and mandibular second premolars.[18] Several studies, however, have reported delayed tooth development in children with clefts.[21–23] Thus, in a child of 6-year old, it may be difficult to determine with confidence agenesis of the second premolar.

With respect to supernumerary teeth, this study showed a prevalence of 17.8% and a maximum value of four supernumerary teeth per individual. A study of Brazilian patients with clefts aged 12 to 45 years reported a lower prevalence. The same study reported that the frequency of supernumerary teeth was higher in patients with bilateral complete CLP.[24] In another study of a sample of 90 patients aged 4–20 years, affected by isolated CL, a higher frequency (30%) was reported for supernumerary teeth in the incisor region.[25]

Peg-shaped teeth were recorded in 7.5% of cleft patients in the present study, predominantly in the maxilla. A higher frequency was found for the left lateral incisors, presumably because of the predominance of left-sided CL in the study population. Paranaiba et al., found a frequency of 8.1% for peg-shaped teeth in a group of cleft patients in Brazil.[8] In accordance with the present study, a frequency of 4.4% of peg-shaped teeth has been reported in a previous investigation.[26] Al-Jamal et al., however, reported a higher frequency.[27] The discrepancy may suggest large geographic variations, but it should also be noted that direct comparison of study findings is complicated by lack of consistency of classification in the different studies.

Oral and facial clefts enhanced the possibility of developing occlusal disorders and thereby increase the orthodontic treatment needs that require a multidisciplinary approach.[28] More than half of the subjects in the present study exhibited one or more occlusion disorders. Angle Class III occlusion with a negative overjet was observed more frequently among CLP patients. In 2002, Baek et al., reported that 84.6% of 39 children in the mixed dentition stage had an Angle class III occlusion.[11] The agenesis in the upper jaw was considered as one of the contributing factors for maxillary hypoplasia and the consequent Angle Class III occlusion.[28–33] In the present study, an Angle Class II occlusion with a positive overjet was found in fewer than ten percent of the study population, this finding is in accordance with previous reports.[34,35] In the present study, unilateral crossbite occurred more frequently than bilateral crossbite and both types of occlusion were more common in CLP patients. Crossbite in the study population exceeded the average for the general population.[36] The same tendency has been reported previously, however, higher frequencies of crossbite and open bite have been reported among patients with clefts.[9,10] Reiser et al., found that crossbite was more frequent in 5-year-old patients with unilateral CLP than in those with CP.[37] The subjects of the present study had a lower overall incidence of open bite than reported in non-cleft subjects of a previous study.[36] In contrast, Chopra et al., reported that anterior open bite and increased overjet were more prevalent among children with clefts.[38]

Interestingly, the present study revealed an almost two folds chance to get Class III occlusion if patients with CLP have agenesis suggesting reduced growth of the maxilla. In contrast patients with supernumerary teeth presented low chance to get Class III occlusion suggesting an enhanced growth of tissue supporting the teeth.

One of the limitations of the present study is the long period for data collection, where different examiners were involved. Since no calibration for the examiners was performed this could affect the accuracy of parameters used for determination of malocclusion. Furthermore, the reports with regards to agenesis and other reported dental anomalies in the present study have to be interpreted carefully since some teeth might be delayed in the development and eruption. The authors however believed that all possible previous errors and or misdiagnosis were corrected in the follow-up control visits since all patients belonging to the CLP team in Bergen are regularly followed up until the age of 16-year old.

Conclusions

There is fluctuation on the prevalence of different cleft types through the years. The findings supported the hypothesis that the distribution of dental anomalies and occlusal disorders varied among patients with CL, CP and CLP. In patients with cleft, there is a twofold chance to get Class III malocclusion in the presence of agenesis. These findings might be of relevance in clinical treatment planning of patients with CL/P.

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Disclosure statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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