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The prevalence of various dental characteristics in the primary and mixed dentition in patients born with non-syndromic unilateral cleft lip with or without cleft palate

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SUMMARY This retrospective study was carried out on consecutively collected dental casts, x-rays, and photos of 129 Swedish children who had been born with non-syndromic unilateral (U) cleft lip (CL), cleft lip and alveolus (CLA), or cleft lip and palate (CLP). The following dental characteristics were investigated in the primary and permanent dentitions: 1. the presence, eruption, position, and shape of the lateral incisor; 2. the prevalence of rotation and enamel hypoplasia of the permanent central incisor; 3. the occurrence of hypodontia outside the cleft region; and 4. the transition from the primary to the succeeding permanent lateral incisor in the cleft region.

Patients with clefts involving the palate (UCLP) exhibited a high frequency of hypodontia. In patients with clefts, which did not include the palate, malformed lateral incisors were a common finding. In the primary and permanent dentition, the lateral incisor had erupted distal to the cleft in 31.8 and 24.8 per cent of the UCLA and UCLP patients, respectively. No significant pattern was seen regarding the transition from the primary to the succeeding permanent lateral incisor ($P = 0.15$). The central incisor was rotated in 55 per cent of the patients and this positional deviation was particularly frequent in cases where the lateral incisor was missing in the premaxilla ($P < 0.05$). Hypodontia outside the cleft region was recorded in 15.5 per cent of the patients. Patients with UCLP had more often crossbite than patients with a UCL or a UCLA phenotype ($P < 0.001$).

Introduction

Clefts of the upper lip (CL), with or without cleft alveolus (CLA) and palate (CLP), or cleft palate only (CP), are among the most common congenital human malformations, showing a mean incidence of 1.7/1000 live births in Stockholm (Milerad *et al.*, 1997). Failure of fusion of the maxillary and medial nasal prominences results in cleft lip of varying extent, unilaterally or bilaterally. By the 7th week *in utero*, the palatal shelves elevate. This is followed by adherence of the medial edge epithelia (MEE) of the opposing palatal shelves. During fusion of the palatal shelves apoptosis of the MEE takes place (Rice, 2005).

The regulatory mechanism for tooth development is based on a communication between the epithelium and the mesenchyme. The mechanism is composed mainly of secreted signal molecules and growth factors (Thesleff, 2006). The dental lamina elongates from the enamel organ of the deciduous tooth and is activated at a later stage to generate the formation of the replacement tooth. The molecular mechanisms, which are responsible for the growth of the dental lamina and the initiation of the development of the replacement tooth, may be regulated independently (Järvinen *et al.*, 2009). The formation of a deciduous tooth, therefore, may not be a prerequisite for the development of the replacement tooth (Casal *et al.*, 2007).

In the literature, there are many synonyms for missing teeth; the term hypodontia has been chosen in this article. The frequency of hypodontia in the permanent dentition is 7.4 per cent in Swedish school children between the ages 7 and 9 years. The frequency of supernumerary teeth varies between 1.5 and 1.9 per cent (Bergström, 1977; Bäckman and Wahlin, 2001).

In a normal population, the supernumerary teeth in the maxilla are usually located in the central incisor area (mesiodens) (Hurlen and Humerfelt, 1985). In a population with clefts, an additional lateral incisor is the most frequent supernumerary tooth (Ringkvist and Thilander, 1969). Furthermore, in the cleft area, most developmental dental irregularities are related to the upper lateral incisor, both in the primary and in the permanent dentitions (Shapira *et al.*, 2000). In the permanent dentition of patients with unilateral (U) clefts, frequencies of 20 per cent of supernumerary

teeth and 43.6 per cent of hypodontia of the lateral incisor have been recorded in the cleft area (Hellquist *et al.*, 1979). It has also been shown that missing teeth are more commonly found in non-syndromic (NS) cleft patients (CLP, CLA, and CP) and their siblings without cleft than in a non-cleft control group. This indicates that clefts and hypodontia to some extent might have a common genetic background (Eerens *et al.*, 2001). It has recently been suggested that the *IRF6* gene, previously reported as being responsible for the Van der Woude syndrome (VWS) (Kondo *et al.*, 2002) and associated with NS oral clefts (Zucchero *et al.*, 2004), might also be involved in the development of isolated hypodontia (Vieira *et al.*, 2008).

Aim

The aim of this study was to investigate a number of dental characteristics in Swedish children born with NS unilateral (U) cleft lip (CL), cleft lip and alveolus (ULA), and cleft lip and palate (CLP). The following traits were recorded: the developmental pattern of the lateral incisor; the presence, position, and shape of the lateral incisor in the primary and permanent dentition; rotation and enamel hypoplasia of the permanent central incisor; hypodontia outside the cleft region; and the transverse occlusal relation of the buccal segments on the cleft versus the non-cleft side. In addition, the transition from the primary to the succeeding permanent lateral incisor in the cleft region was investigated.

Subjects and methods

Subjects

A consecutive series of 129 Caucasian children born with NS UCL, UCLA or UCLP between 1986 and 1997 and treated by the Stockholm Craniofacial Team, were included in this retrospective study. The distinction between UCL and UCLA patients was based on information obtained from the case records. Patients in whom a bone graft to the alveolus had been needed for the erupting tooth formed the UCLA group, whereas those who did not need this treatment were classified as being UCL patients.

The material consisted of radiographs, dental casts, and photos taken at the ages of 5, 7, and 10 years. For the analysis of the transverse occlusion, 30 patients had to be excluded, 29 due to incomplete dental casts and one due to a bilateral scissors bite.

An approval for this study has been obtained from the ethical committee in Stockholm (2006/1243-31/4).

Methods

In the primary dentition, the developmental pattern of the lateral incisor in the cleft region was studied and subgroups based on the presence, position, and shape of this tooth were formed.

In the mixed dentition, the developmental pattern of the permanent lateral incisor in the cleft region was also studied and subgroups based on the presence, position, and shape of this tooth were formed. As regards the permanent central incisor on the cleft side, the presence of rotations and enamel hypoplasia was recorded. Hypodontia outside the cleft region was also recorded.

Dental casts and photos of the mixed dentition were used to study and compare the transverse and sagittal occlusion in the buccal segments on the cleft and the non-cleft side. A modified index by Huddart and Bodenham (1972) was applied in the UCLP patients in order to verify our findings. Mossey *et al.* (2003) have modified the Huddart and Bodenham index for use in the permanent dentition. However, in contrast to Huddart and Bodenham (1972) and Mossey *et al.* (2003), only positive index values were used in the present study. Consequently, the index value was recorded as zero in cases of normal occlusal relation.

The developmental pattern of the lateral incisor in the cleft area during the transition from the primary to permanent dentition was studied in all patients. The findings in the UCLA and UCLP groups were compared with previous results in this field that have been reported by Ranta (1971) and Tsai *et al.* (1998).

Statistics

The Statistical software (version 8; StatSoft, Inc., USA) was used for analysis of the recorded data. Differences in proportion of the various characteristics recorded in the three groups were tested for statistical significance using the chi-square test for one sample (Table 1) and Pearson's chi-square test (Tables 1–9). In Tables 11 and 13, differences were analysed with the McNemar and Fisher tests (Table 11) and the Sign test (Table 13). The level of significance was $P < 0.05$.

Results

The distribution of different types of cleft in relation to sex and side of the clefts are presented in Table 1. In the total, material clefts were significantly more common in boys than in girls ($P < 0.001$), and the clefts were mainly found on the left side ($P < 0.001$).

Primary lateral incisors

The positions of the primary lateral incisors in relation to the types of cleft are presented in Table 2. In the total sample, 31.8 per cent of the primary lateral incisors had erupted distal and 1.6 per cent had erupted mesial to the cleft. Supernumerary lateral incisors were recorded in 34.9 per cent and hypodontia in 12.4 per cent of the patients. The frequencies of hypodontia and supernumerary teeth in

Table 1 Prevalence of different cleft types in relation to gender and cleft side. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Diagnosis	Patients		Gender		Side	
	n	%	Female, %	Male, %	Left, %	Right, %
UCLP	67	52.0	14.0	38.0	39.0	13.0
UCLA	15	11.6	3.1	8.5	9.3	2.0
UCL	47	36.4	10.1	26.4	27.1	9.3
Total	129	100.0	27.0	72.9***	76.0***	24.0

***$P < 0.001$.

Table 2 Position and prevalence of the primary lateral incisor, supernumerary lateral incisors, and hypodontia in the region of the cleft in 129 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Cleft type	Mesial		Distal		Supernumerary		Hypodontia		Normal		Total row	
	n	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%
UCLP	2	3.0	33	49.2	17***	25.4	15	22.4	0	0	67	52.0
UCLA	0	0	8	53.3	7***	46.7	0	0	0	0	15	11.6
UCL	0	0	0	0	21***	44.7	1	2.1	25	53.3	47	36.4
Total	2		41		45		16		25		129	100.0
Total %		1.6		31.8		34.9		12.4		19.4		100.0

***$P < 0.001$ more common in UCL and UCLA compared to UCLP.

Table 3 Prevalence of variations in size and shape of the primary lateral incisor in the cleft region related to cleft type in 113 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Cleft type	Macrodontia		Malformed		Normal		Total row	
	n	%	*n*	%	*n*	%	*n*	%
UCLP	7	13.5	11	21.2	34	65.4	52	46.0
UCLA	3	20.0	4	26.7	8	53.3	15	13.3
UCL	14	30.4	9	19.6	23	50.0	46	40.7
Total	24		24		65		113	
Total%		21.2		21.2		57.6		100.0

the UCLP patients were 22.4 and 25.4 per cent, respectively. In the patients with UCL, 2.1 per cent of hypodontia and 44.7 per cent of supernumerary laterals were recorded. When the patients with the diagnoses UCL and UCLA were combined into one group, the prevalence of supernumerary lateral incisors in that group was significantly greater than in the UCLP group ($P < 0.001$).

The occurrence of variations in size and shape of the primary lateral incisor is shown in Table 3. The percentage of patients with such morphological discrepancies was 21.2 per cent, both in respect of macrodontia and in respect of malformation.

Permanent lateral incisors

The positions of the permanent lateral incisors in relation to the types of cleft are presented in Table 4. In the total sample, 24.8 per cent of the permanent lateral incisors had erupted distal and 4.6 per cent had erupted mesial to the cleft. Supernumerary lateral incisors were recorded in 17.0 per cent and hypodontia in 29.5 per cent of the patients.

In the UCLP group, 43.3 per cent of the patients exhibited hypodontia and 14.9 per cent had a supernumerary lateral incisor. In the UCL patients, 66.0 per cent had one normally

positioned lateral incisor and supernumerary lateral incisors was present in 21.3 per cent of the cases.

The prevalence of deviations in size and shape or hypodontia of the lateral incisor in relation to the types of cleft is presented in Table 5. Only 26.4 per cent of the patients exhibited normal shape of this tooth in the cleft area. Microdontia was recorded in 12.4 per cent and malformation in 3.9 per cent of the patients. The combination malformation/pegshape showed a frequency of 4.6 per cent. Pegshaped laterals were found in 23.3 per cent of the total sample. In the separate patient groups, pegshaped teeth were recorded in 53.2 per cent of the UCL patients and in 6.0 and 6.7 per cent of the UCLP and UCLA patients, respectively. The occurrence of the various malformations differed significantly between the three patient groups ($P < 0.001$).

Permanent central incisor

The prevalence of central incisor rotations and enamel–malformations are presented in Tables 6–8. Incisor rotation was recorded in 55.0 per cent of the patients and most common was the mesio-palatal type of rotation (47.3 per cent, Table 6). In the UCLP patients, rotated central incisors were found in 83.6 per cent of the cases and 74.6 per cent of these were mesio-palatal rotations (Figures 1 and 2). In cases where the permanent lateral incisor was absent in the premaxilla (positioned distal to the cleft or missing), 78.6 per cent of rotations of the central incisor were recorded (Table 7, $P < 0.05$).

The central incisor had enamel opacities or enamel hypoplasia in 48.1 per cent of the patients in the total sample. In the UCLA patient group, such defects were found in 80 per cent of the cases (Table 8, Figures 3 and 4).

Table 4 Position and prevalence of the permanent lateral incisor, supernumeray lateral incisors, and hypodontia in the region of the cleft in 129 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Cleft type	Mesial		Distal		Supernumerary		Hypodontia		Normal		Total row	
	n	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%
UCLP	5	7.5	23	34.3	10	14.9	29	43.3	0	0	67	52.0
UCLA	1	6.7	9	60.0	2	13.3	3	20.0	0	0	15	11.6
UCL	0	0	0	0	10	21.3	6	12.8	31	66.0	47	36.4
Total	6		32		22		38		31		129	100.0
Total%		4.6		24.8		17.0		29.5		24.0		100.0

Table 5 Prevalence of deviations in size and shape or hypodontia of the permanent lateral incisor in the cleft region related to cleft type in 129 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Cleft type	Pegshape		Malformed		Microdontia		Peg shape and malformed		Total abnormal shape		Hypodontia		Normal		Total row	
	n	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%
UCLP	4	6.0	0	0	8	11.9	4	6.0	16	23.9	29	43.3	22	32.8	67	52.0
UCLA	1	6.7	3	20.0	0	0	1	6.7	5	33.3	3	20.0	7	46.7	15	11.6
UCL	25	53.2	2	4.3	8	17.0	1	2.1	36	76.6	6	12.8	5	10.6	47	36.4
Total	30		5		16		6		57		38		34		129	
Total%		23.3		3.9		12.4		4.6		44.2		29.5		26.4		100.0

$P < 0.001$ significant difference between the cleft types.

Table 6 Prevalence of central incisor rotation related to cleft type in 129 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Central incisor rotation							
Cleft type	*n*	Mesio-palatal		Mesio-buccal		No rotation	
		n	%	*n*	%	*n*	%
UCLP	67	50	74.6	6	9.0	11	16.4
UCLA	15	9	60.0	1	6.7	5	33.3
UCL	47	2	4.2	3	6.4	42	89.4
Total	129	61	47.3	10	7.7	58	45.0

Table 7 Prevalence of central incisor rotation in relation to the position of the lateral incisor on the cleft side in 129 patients with UCL, UCLA and UCLP. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

		Central incisor rotation					
	Total, *n*	Mesio-palatal		Mesio-buccal		No rotation	
		n	%	*n*	%	*n*	%
Perm lateral incisor							
Mesial	6	6	4.6	0	0	0	0
Distal	32	22	17.0	4	3.1	6	4.6
Hypodontia	38	26	20.2	3	2.3	9	7.0
Supernumerary	22	5	3.9	2	1.6	13	10.1
Normal	31	2	1.6	1	0.8	30	26.3
Total	129	61	47.3	10	7.7	58	45.0

Distal placement/hypodontia combined compare to mesial/supernumerary, i.e. when no lateral incisor is present in the premaxilla the central incisor is more often rotated ($P < 0.05$); % express the percentage of the total material.

Table 8 Prevalence of enamel malformation on the central incisor related to cleft type in 129 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Cleft type	Central incisor enamel							
	Hypoplasia		Opacity		Normal		Total row	
	n	%	*n*	%	*n*	%	*n*	%
UCLP	19	28.4	15	22.4	33	49.2	67	52.0
UCLA	9	60.0	3	20.0	3	20.0	15	11.6
UCL	6	12.8	10	21.3	31	66.0	47	36.4
Total	34		28		67		129	
Total%		26.4		21.7		51.9		100.0

Figure 1 Mesio-palatal rotation of 21.

The majority of those who exhibited abnormal shape of the permanent lateral incisor mostly had central incisors with normal enamel ($P < 0.01$). Furthermore, hypodontia of the permanent lateral incisor was observed in 14 of the patients whose central incisor exhibited enamel hypoplasia (Table 9).

Figure 2 Mesio-palatal rotation of 21, aplasia of 22, and unilateral crossbite.

Figure 3 Enamel opacity on 21.

Figure 4 Enamel hypoplasia on 21.

Hypodontia outside the region of the cleft

Hypodontia outside the region of the cleft was diagnosed in 15.5 per cent (20 individuals) of the patients. Table 10 illustrates how this condition was related to type of cleft, side of the cleft, sex, and presence or absence of the lateral incisor in the cleft area. In 50.0 per cent of these patients, hypodontia was recorded, both inside and outside the cleft area. In the total material, hypodontia of the permanent lateral incisor combined with hypodontia outside the cleft area was found in 37.0 per cent of the patients.

Table 9 Prevalence of central incisor enamel malformation in relation to the lateral incisor shape in 91 patients and in relation to the prevalence of hypodontia in 38 patients with UCL, UCLA and UCLP. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Central incisor enamel	Lateral incisor shape														Lateral incisor absence
	Pegshape		Malformed		Microdontia		Pegshape + malformed		Total abnormal shape		Normal		Total row		Hypodontia
	n	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%	
Normal	19	38.0	2	4.0	11	22.0	5	5.0	37**	74.0	13	26.0	50**	55.0	17
Opacity	8	38.0	1	4.8	3	14.3	0	0	12	57.1	9	42.6	21	23.0	7
Hypoplasia	3	15.0	2	10.0	2	10	1	5.0	8	40.0	12	60.0	20	22.0	14
Total	30		5		16		6		57		34		91		38
Total%	33.3		5.5		17.6		6.6		62.6		37.4		100.1		

Table 10 Hypodontia found outside the region of the cleft, in relation to cleft type and side of cleft, sex, and presence of a lateral incisor in the cleft area in 15.5 per cent of the patients (20 of the 129 patients). F, female; M, male; UCL, unilateral cleft lip; UCLP, unilateral cleft lip and palate.

Cleft type	Sex	Cleft side	Lateral incisor, cleft region	Hypodontia outside the region of the cleft
UCLP	F	left	—	12
UCLP	M	left	—	12
UCLP	M	left	—	45
UCLP	F	left	—	35
UCLP	F	left	—	47
UCLP	F	right	—	31, 41
UCLP	F	right	—	25, 35
UCLP	M	right	12, 12	25
UCLP	F	right	12, 12	15, 25, 35, 45
UCLP	M	right	12	44, 45
UCLP	F	left	22	35
UCLP	M	right	12	14, 24, 34, 44
UCLP	M	left	22	24, 25
UCLP	M	right	12	24
UCL	M	right	—	45
UCL	F	right	—	15, 25
UCL	M	left	—	12
UCL	M	right	12	22
UCL	F	left	22	12
UCL	M	right	12	45

Occlusion

Transverse relation. The transverse occlusal relation of the buccal segments in 99 patients with different types of clefts is presented in Table 11. In comparison with the unaffected side, significantly more crossbites were recorded on the cleft side ($P < 0.001$). Also, between the different cleft groups, the transverse relations differed significantly, with the majority of crossbites observed in the UCLP group ($P < 0.001$; Table 11). The modified Huddart and Bodenham index showed a mean value of 3.9 on the cleft side as compared with 0.6 on the non-cleft side (Table 12).

Frontal relation. In the permanent dentition, an anterior crossbite was present in 39.1 per cent with UCLP (Table 12). The transition from the primary to the succeeding permanent lateral incisor.

The eruption pattern for the lateral incisor in the cleft region in the total sample, following the same patients is shown in Table 13. In 21 of 41 patients, in whom the primary lateral incisor had erupted distal to the cleft, the permanent lateral incisor also erupted in a distal position (51.2 per cent). A primary lateral incisor in a position mesial to the cleft was recorded in two patients. Of the corresponding permanent lateral incisors, one erupted mesial and the other distal to the cleft. In 81.2 per cent (13/16) of the cases, hypodontia of the primary lateral incisor was followed by hypodontia of the permanent lateral incisor. Primary supernumerary lateral incisors were succeeded by the same eruption pattern in 35.6 per cent (16/45). No significant pattern was seen as regards the transition from the primary to the succeeding permanent lateral incisor ($P = 0.15$).

Our results of the eruption patterns for the lateral incisor compared to Ranta (1971) and Tsai *et al.* (1998) are shown in Table 14. In all studies, the distal position was the most common in the primary dentition. In our study, in the permanent dentition, the distal position and hypodontia had the same frequency.

Discussion

NS CLP is a malformation with a multifactorial cause in which both genetic and environmental factors determine the probability to develop the anomaly. These malformations of multifactorial origin show gender discrepancies. The

Table 11 The transverse occlusal relation of the buccal segments in relation to type of cleft in 99 patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Cleft type	Cleft side				Non-cleft side				Total *n*
	No crossbite		Crossbite		No crossbite		Crossbite		
	n	%	*n*	%	*n*	%	*n*	%	
UCLP	9	14.0	55	86.0	54	84.0	10	15.6	64
UCLA	6	75.0	2	25.0	8	100.0	0	0	8
UCL	25	93.0	2	7.4	26	96.5	1	3.4	27
Total	40		59		88		11		99
Total %	40.4		60.6		88.9		11.1		

Table 12 The transverse buccal and frontal occlusal relation in cleft lip and palate patients (n = 64) evaluated with a modified index by Huddart and Bodenham.

	n	Score	Total score	Mean score
Frontal relation				
Normal	18	0		
Edge to edge	21	21		
Anterior crossbite	25	50		
			71	1.1
Crossbite cleft side				
Cr1	26	52		
Cr2	11	55		
Cr3	18	144		
			251	3.9
Crossbite non-cleft side				
Cr1	6	12		
Cr2	2	10		
Cr3	2	16		
			38	0.6

Cr1 canine crossbite = 2 p; Cr2 canine crossbite, premolar/primary molars, and molars edge to edge = 2 + 1 + 1 + 1 = 5 p; Cr3 canine to molar crossbite = 2 + 2 + 2 + 2 = 8 p; score: 0 = normal relation; 1 = edge to edge transverse relation; 2 = crossbite; two patients had incomplete records and were excluded; one patient had a scissors bite and was excluded.

majority of the patients in the present material were males, and this is in accordance with the results presented by Böhn (1963). Furthermore, cleft lip (CL) is more common in males, whereas isolated cleft palate (CP) is more often diagnosed in females (Shapira *et al.*, 1999). The cleft was more common on the left side, which is in line with data previously published by Shapira *et al.* (1999).

This investigation was a retrospective study and the patients were consecutively included, which may explain the variation in number of patients within each cleft group. In patients with an alveolar cleft (UCLA and UCLP), bone grafting is performed after the development of the permanent tooth bud but before the eruption of the permanent tooth in the cleft area. This is carried out in order to provide bone support for the erupting tooth. The distinction between UCL and UCLA is essential as a cleft involving the alveolus may alter the developmental condition for the tooth bud.

Lateral incisor

In a material of healthy macaque, foetuses Wei *et al.* (2000) could show that the lateral incisors undergoes a complex positional shift (mainly medial) during the pre- and post-natal path of development and are finally located medial to the premaxillary/maxillary suture. Furthermore, in another material consisting of macaque foetuses with induced CLP, they could demonstrate that the lateral incisor was located distal to the alveolar cleft and did not shift medial to the premaxillary/maxillary suture. In the light of these facts, it was not surprising to find that the majority of both the primary and the permanent lateral incisors had erupted distal to the cleft in the present material in the UCLA and UCLP group.

A supernumerary tooth in the cleft region was found more frequently in the primary than in the mixed dentition. A similar result has previously been presented by Ringkvist and Thilander (1969). Supernumerary teeth are relatively often found in the primary dentition, whereas hypodontia is more frequent in the permanent dentition. A hypothesis by Hovorakova *et al.* (2006) suggests that formation of supernumerary teeth is the result of the non-fusion of the medial nasal and maxillary processes which also leads to separation of the dental epithelia (which normally fuses 4–6 days later than the rest of the facial processes), thus giving rise to two lateral incisors, one on each side of the cleft.

A greater number of supernumerary lateral incisors were recorded in the primary dentitions of the UCL and UCLA groups than in those of the UCLP group. Hansen and Mehdinia (2002) reported a frequency of 73 per cent of supernumerary primary and permanent lateral incisors in patients with UCL. The corresponding frequency in the present material was 66.0 per cent.

The frequency of hypodontia in the present UCLP patients' mixed dentitions is in line with data from a similar study by Tortora *et al.* (2008). In addition, they reported that

Table 13 Distribution of the transition from the primary to the succeeding permanent lateral incisor in the region of the cleft in 129 UCL, UCLA and UCLP patients. UCL, unilateral cleft lip; UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

Primary lateral incisor position	Permanent lateral incisor											
	Mesial		Distal		Supernumerary		Hypodontia		Normal		Primary lateral incisor	
	n	%	*n*	%	*n*	%	*n*	%	*n*	%	*n*	%
Mesial	1	0.8	1	0.8	0	0	0	0	0	0	2	1.6
Distal	3	2.3	21	16.3	3	2.3	14	10.8	0	0	41	31.8
Hypodontia	1	0.8	1	0.8	1	0.8	13	10.1	0	0	16	12.5
Supernumerary	1	0.8	9	7.0	16	12.4	7	5.4	12	9.3	45	34.9
Normal	0	0	0	0	2	1.6	4	3.1	19	14.7	25	19.4
Total permanent lateral incisor	6	4.6	32	24.8	22	17.0	38	29.5	31	24.0	129	100.0

%, percentage of the total material.

Table 14. The prevalence of four eruption patterns for the primary and permanent lateral incisor in 82 patients with UCLA or UCLP also showing evaluations made by Ranta (1971) and Tsai *et al.* (1998). UCLA, unilateral cleft lip and alveolus; UCLP, unilateral cleft lip and palate.

	Present study, *n* = 82 (%)	Ranta (1971), *n* = 47 (%)	Tsai *et al.* (1998), *n* = 91 (%)
Primary lateral incisor erupting in segment			
Mesial	2.4	17.0	2.2
Distal	50.0	48.9	82.4
Supernumerary	29.3	27.7	5.5
Hypodontia	18.3	6.4	9.9
Permanent lateral incisor erupting in segment			
	n = 82	*n* = 83	*n* = 137
Mesial	7.3	7.2	1.5
Distal	39.0	22.9	46.0
Supernumerary	14.6	31.3	0.7
Hypodontia	39.0	38.6	51.8

this diagnosis also applied to lateral incisors on the contralateral side in 6.1 per cent of the cases. In the present material, the corresponding frequency was 3.1 per cent. A UCL and palate in combination with bilaterally missing lateral incisors might possibly indicate that a bilateral disturbance in the craniofacial development has occurred, however less severe on the non-cleft side (Letra *et al.*, 2007).

Disturbed tooth formation could be the result of the presence of a cleft as this anomaly may adversely alter the tissue anatomy and the blood and nerve supply in the area of the developing tooth bud (Kjaer *et al.*, 1994). In the primary dentitions of the present patient groups, some of the laterals were macrodontic, whereas in the permanent dentitions, the lateral incisors were often microdontic or pegshaped. In order to diagnose shape anomalies, the teeth were compared qualitatively with their contralateral or the neighbouring teeth. A total of 44.2 per cent of the permanent lateral incisors exhibited abnormal shape, and pegshape was the most common abnormality, which is in agreement with the results reported by Rawashdeh and Abu Sirdaneh (2009). The majority of the pegshaped permanent lateral incisors were associated with the UCL phenotype. In patients with UCLP, hypodontia and normal tooth shape were more common features.

The central incisor

Patients, in whom the permanent lateral incisor was missing in the premaxilla, exhibited significantly more rotations of the central incisor. To some extent, the higher frequency of rotations of the central incisor, recorded in patients in whom the lateral incisor was positioned in the distal segment or missing, could be explained by the fact that the amount of alveolar bone distal to the root of the central incisor in such cases is reduced, and this situation exists already at the stage of eruption of the tooth. However, Ranta (1971) who also studied the occurrence of rotated central incisors in UCLP children did not find any relation between such rotations and presence or absence of the lateral incisor.

Enamel opacity and/or enamel hypoplasia may be caused by developmental or pathological disturbances during amelogenesis or by a mechanical trauma during enamel maturation. Disturbed enamel formation in the permanent teeth is most frequently seen in the central incisor on the cleft side (Maciel *et al.*, 2005). In the present material, enamel opacity or enamel hypoplasia was recorded in 48.1 per cent of the patients. Hellquist *et al.* (1979) reported a similar result.

Hypodontia outside the region of the cleft

There were no sex differences in respect of hypodontia of permanent teeth outside the region of the cleft. Ten of the 20

patients with hypodontia outside the cleft region also had a lateral incisor missing in the cleft area. Hypodontia in and outside the cleft area in the total material was 37 per cent. Shapira *et al.* (2000) reported a higher incidence of hypodontia in and outside the region of the cleft in their patients (77 per cent). Hypodontia outside the cleft area was most common in the UCLP patients. An interpretation of this result could be that the frequency of hypodontia outside the region of the cleft is related to the degree to which the palate is affected. In patients from Stockholm born with NSCP a significant relation between the length of the cleft and the prevalence of hypodontia was found. The higher the number of missing second premolars, the more extended was the length of the cleft (Karsten *et al.*, 2005).

Occlusion

Transverse relation. The modified Huddart and Bodenham index with only positive values is used in the Swedish national quality register for cleft patients. In the present UCLP patients, the frequency of lateral crossbites was higher on the cleft than the non-cleft side. This suggests that the development of crossbite could mainly be attributed to the cleft itself or to the surgical repair of the palate. A surgically repaired complete UCLP results in a deformation of the alveolus and palate (DiBiase *et al.*, 2002) and as a consequence of this a crossbite with all teeth involved on the cleft side may develop. A cleft solely affecting the lip and alveolus, results in a limited deformation and a crossbite in these cases usually involves only the teeth in the cleft area. Patients with a cleft only in the lip have fewer crossbites (Garrahy *et al.*, 2005).

Frontal relation. The scar formation after the primary lip and/or palate surgery may restrain the anterior growth of the maxilla. A developmental maxillary hypoplasia could also be the consequence of the cleft anomaly itself. As regards occlusion, the maxillary retrusion often results in anterior crossbite. In the present material, this malocclusion was registered in the primary dentition of 2 patients and in the mixed dentition of 25 patients. Holst *et al.* (2009) reported that maxillary retrusion becomes more noticeable in CLP patients with increasing age.

The transition from the primary to the succeeding permanent lateral incisor. Regarding the transition from the primary to the succeeding permanent lateral incisor, in the cleft area, no clear pattern could be shown. This finding is in accordance with previous results presented by Tsai *et al.* (1998).

Previous studies of the lateral incisor in the cleft area have been limited to patients with UCLA and UCLP (Ranta, 1971; Tsai *et al.*, 1998). In the present material, when excluding the UCL patients (n = 47), distally positioned lateral incisors in the remaining UCLA and UCLP patients were recorded with a frequency of 50.0 and 39.0 per cent, in the primary and permanent dentitions, respectively. High frequencies of distally positioned lateral incisors in the cleft region have previously been reported by Ranta (1971) and Tsai *et al.* (1998).

Information regarding dental anomalies is an important tool in genetic analyses in the field (Menezes and Vieira, 2008). A significantly higher frequency of dental anomalies has been found in non-cleft siblings of patients with clefts than in a non-cleft population (Schroeder and Green, 1975).

In future investigations, dental anomalies in patients who are born with CL ± P (cleft lip and/or palate) or VWS will be studied with focus on the influence of the *IRF6* gene.

Conclusions

The different cleft phenotypes studied exhibited differences in dental expression. In patients with clefts including the palate (UCLP), hypodontia was a frequent finding. Clefts, which did not include the palate, were associated with a high frequency of malformed lateral incisors. Most of the existing lateral incisors were positioned distal to the cleft. No significant pattern was seen regarding the transition from the primary to the succeeding permanent lateral incisor, in the cleft area (P = 0.15). Malformed lateral incisors were often found in patients where the central incisor had normal enamel ($P < 0.01$). A central incisor rotation was particularly frequent in cases where the lateral incisor was missing in the premaxilla ($P < 0.05$). Patients with UCLP exhibited more often crossbites than patients with a UCL or a UCLA phenotype ($P < 0.001$). Moreover, in patients with UCLP, crossbite was more common on the cleft side as compared to the non-cleft side.

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