Treatment of Dental Anomalies in Children With Complete Unilateral Cleft Lip and Palate at SickKids Hospital, Toronto

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Objective: **To quantify dental anomalies in permanent dentition associated with complete unilateral cleft lip and palate and to survey treatment modalities used to address these problems.**

Method: **Retrospective study of 116 children with complete unilateral cleft lip and palate treated at SickKids since birth. Presence and morphology of lateral incisors and second premolars were determined. Orthodontic, surgical, and/or prosthetic procedures were analyzed.**

Results: **The cleft-side lateral incisor was absent in 93.1% of finished cases. The lateral incisor mesial to the cleft was present in 4.3%, absent due to agenesis in 75.9%, and extracted in 19.8% of cases. The lateral distal to the cleft was present in 2.6%, absent due to agenesis in 33.6%, and extracted in 63.8% of cases. Of 105 lateral incisors, only one had normal morphology. Noncleft-side lateral incisors were absent in 16% of finished cases. Absence was due to agenesis in 12.1% of cases and extraction in 4.3%. When the lateral incisor was missing, closure of the dental space occurred by orthodontic tooth movement after alveolar bone grafting (45%); surgical closure with simultaneous alveolar bone grafting (35%); prosthetic closure (17%); and 3% were failures. Agenesis of premolars occurred in 12.1% of cleft-side and 10.3% of noncleft-side maxillary second premolars.**

Conclusions: **The cleft-side lateral incisor is rarely present at the conclusion of orthodontic and surgical treatment of complete unilateral cleft lip and palate. Often absent due to agenesis, when present it is typically abnormal in size and bone support and is commonly extracted in favor of canine substitution.**

KEY WORDS: *agenesis, complete unilateral cleft lip and palate, lateral incisors*

The dentition of individuals with cleft lip and palate has a greater frequency of anomalies than that found in the noncleft population. These differences more commonly involve the teeth in the vicinity of the cleft. The lateral incisor tooth bud developing in the region of the alveolar cleft is therefore very sensitive to developmental disorders (Olin, 1964; Ranta, 1986; Shapira et al., 2000; Baek and Kim, 2007). The repercussions of the presence of a cleft in the region of the lateral incisor include anomalies in number of teeth (missing or supernumerary), location (mesial or distal to the cleft), shape (pegged or conical), size (microdontic), time of formation and/or eruption, and crown and root malformation (Nagai et al., 1965; Wei et al., 2000). These teeth not only create aesthetic concerns but can also potentially cause functional, periodontal, and restorative problems. The preparation of such small, irregularly shaped teeth for fixed restorations may be difficult. Periodontal problems also may be a concern because these teeth typically have inadequate bony support due to their proximity to the cleft (Shashua and Omnell, 2000).

To our knowledge, there are no detailed clinical studies in the literature surveying orthodontic and surgical treatment rendered at specific institutions for patients with cleft lip and palate.

The purpose of this study was to quantify dental anomalies in the permanent dentition associated with complete unilateral cleft lip and palate (CUCLP) and to survey the treatment modalities being used at SickKids Hospital to address these problems.

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TABLE 1 Development of Cleft-Side Lateral Incisors*

	Lm Agenesis (%)	*Lm Developed (%)*	*Total (%)*
Ld agenesis	26.7	6.9	33.6
Ld developed	49.1	17.2	66.3
Total	75.8	24.1	100.0

* Ld = lateral incisor distal to the cleft; Lm = lateral incisor mesial to the cleft.

TABLE 2 The Lateral Incisors at Conclusion of Treatment

	Lateral Mesial to the Cleft (%)	*Lateral Distal to the Cleft (%)*	*Lateral on Noncleft Side (%)*
Present	5 (4.3)	3 (2.6)	97 (83.6)
Missing—agenesis	88 (75.9)	39 (33.6)	14 (12.1)
Missing—extracted	23 (19.8)	74 (63.8)	5 (4.3)

Sample and Methods

This was a retrospective study of 116 children (68 boys and 48 girls) with nonsyndromic CUCLP, who completed both orthodontic and surgical treatment at SickKids Hospital, Toronto, Canada, between 1990 and 2006. Nonsyndromic patients were selected to avoid possible dental anomalies associated with other syndromes. Only patients who received their complete orthodontic treatment at SickKids and who had adequate records for full evaluation were included in this study. All subjects were born between 1975 and 1993. Approval from the SickKids Research Ethics Board was obtained prior to beginning data collection.

All subjects had pre- and posttreatment records that permitted tooth identification and morphological description of all permanent teeth excluding third molars. Pretreatment records included any records that were necessary to determine the presence and history of the teeth in question (panoramic, occlusal, periapical, and cephalometric radiographs, as well as clinical photographs and orthodontic clinic charts).

For each subject, the presence of a maxillary permanent lateral incisor mesial (Lm) or distal (Ld) to the cleft and its morphology were determined. The tooth distal to the cleft was considered to be a supernumerary lateral incisor (Ld) as per Fishman (1970) and Böhn (1950). Some consider this tooth to be a lateral incisor, but this distinction was not deemed important for this study. Canines were not evaluated in this study in terms of crown and root morphology. Also noted was the presence or absence of the first and second premolar in both maxillary and mandibular arches, on both cleft and noncleft sides. Information was recorded on alveolar bone grafting, orthodontic treatment, orthognathic surgery, and prosthetic treatment, if applicable.

The Lateral Incisors

Using photographs and radiographs, the size and morphology of the lateral incisor crown and root were established by comparison with the noncleft-side central incisor because the cleft-side central and noncleft-side lateral are often of smaller size. Ideally, the mesiodistal width of the maxillary lateral incisor should be about two thirds the width of the central incisor (Kokich, 1993). Exact measurements of the lateral incisors were not recorded in this study; an overall assessment of size and shape was performed with the aid of photos and radiographs. In the current study a lateral incisor was considered small if it was less than 80% of the expected width based on two thirds of the width of the noncleft central incisor. For crown shape, a qualitative designation of normal or abnormal (conical or peg-shaped) was assigned subjectively to each tooth. A qualitative designation also was used for root shape based on the size of the noncleft-side lateral incisor. The noncleft-side lateral incisor was evaluated using a similar protocol.

The Premolars

The first and second premolars in the maxilla and mandible were identified as present, missing (due to agenesis), or extracted (to facilitate orthodontic treatment or due to caries).

Evaluating the Surgical and Prosthetic Treatment

The surgical and prosthetic treatment rendered in each case was recorded. Surgeries included alveolar bone grafting on the cleft side in the region of the lateral incisor or orthognathic surgery involving a single-piece or segmental maxillary osteotomy. If an alveolar bone graft (ABG) was performed, the age of the patient at the time of surgery was noted. If a prosthesis was required in the cleft region to replace the lateral incisor or other missing teeth, this information was also recorded. A case in which a patient did not proceed with prosthetic recommendations for the cleft-side lateral incisor was classified as a failure.

Statistics

A series of chi-square tests, odds ratios, and binomial proportions were conducted on the collected data. A significance level of $p = .01$ was applied to all analyses.

Intraexaminer reliability for identification of missing teeth and size and shape of lateral incisors was conducted by the primary investigator through random selection of a subsample of 20 patients 2 weeks after the initial identification. The same results were found between the two trials, indicating 100% reliability.

Results

Cleft-side and Noncleft-side Lateral Incisors

Tables 1, 2, and 3 summarize the findings of the cleft-side and noncleft-side lateral incisors.

TABLE 3 Morphology of the Lateral Incisors

	Lateral Mesial to the Cleft (%)	*Lateral Distal to the Cleft (%)*	*Lateral on Noncleft Side (%)*
Normal range	1 (3.6)	0 (0.0)	96 (94.1)
Small in size/shape: <80% of normal	27 (96.4)	77 (100.0)	6 (5.9)

There was agenesis in 76% of Lm and 34% of Ld. Agenesis occurred in both Lm and Ld in 27% of cases, and both were present in 17% of cases (Table 1). The Lm was present in 4.3% of finished cases, missing due to agenesis in 75.9%, and extracted to facilitate treatment in 19.8% of cases. The Ld was present in 2.6% of finished cases, missing due to agenesis in 33.6%, and extracted to facilitate treatment in 63.8% of cases. The noncleft-side lateral incisor was present in 83.6% of finished cases, missing due to agenesis in 12.1%, and extracted to facilitate treatment in 4.3% of cases (Table 2). In only 1 of 28 cases (4%) was the Lm normal in size, and in none of the 77 cases was the Ld of normal size. Morphologically normal noncleft-side lateral incisors were present in 94.1% of cases (Table 3).

Premolars

Agenesis occurred in 12.1% of cleft-side maxillary second premolars and 10.3% of noncleft-side maxillary second premolars, which was not significant. Agenesis occurred in 5.2% of cleft-side mandibular second premolars and 4.3% of noncleft-side mandibular second premolars, which was not significant. On the cleft side 6.0% of second premolars and 6.9% of first premolars were extracted to facilitate treatment; whereas, on the noncleft side 6.9% of second premolars and 9.5% of first premolars were extracted for treatment (Table 4).

Odds ratios were calculated to determine if there was an associated agenesis of the cleft-side lateral and second premolar. The odds ratios were 0.4195 ($p = .2620$) and 1.4259 ($p = .5848$), respectively, indicating no significant correlation.

Alveolar Bone Graft

An ABG was placed in 94% of cases. An ABG was done in 50.9% of mixed dentition cases and 38.8% of permanent dentition cases (Table 5).

TABLE 5 Alveolar Bone Graft (ABG)

	No. of Patients (% of Total)
Never had ABG	6 (5.2)
Mixed dentition	59 (50.9)
Before 10 years of age	24 (20.7)
After 10 years of age	35 (30.2)
Permanent dentition	45 (38.8)
In conjunction with orthognathic surgery	38 (32.8)
No other surgery	7 (6.0)
Mixed and permanent dentition	4 (3.5)
Mixed dentition failed, no other ABG	2 (1.7)

Surgery

Orthognathic surgery was performed in 65.5% of cases: 31.0% had one-piece Le Fort I osteotomies and 34.5% had two-piece segmental Le Fort I osteotomies (Table 6).

Prosthodontic Treatment

Lateral incisor tooth replacement was not required for 80.2% of patients because the space in the cleft area was bone grafted and closed by orthodontics (45.7%), or it was closed by a two-piece Le Fort I osteotomy with segment advancement and bone graft (34.5%). A dental space remained open in 20 (17.2%) cases and various prostheses were provided: 12 (10.3%) patients received a single tooth implant to replace the cleft-side lateral incisor; six (5.2%) patients received a removable prosthesis; and two (1.7%) patients received a fixed bridge. Three subjects (2.6%) refused the recommended prosthodontic treatment and were classed as failures (Table 7).

Comparing Results Between Different Time Periods

The subjects were further divided into two groups based on whether they completed their treatment in the 1990s (n = 42) or the 2000s (n = 74). Neither surgical nor prosthetic treatment showed a change in the type of treatment performed. There were no statistical differences in the treatment rendered between the two time periods (Table 8).

Transpositions

Ten cases of transpositions of maxillary canines and first premolars were found in this study (8.6% of cases), all on the cleft side.

TABLE 4 Maxillary and Mandibular Premolar Evaluation*

	mxC5 (%)	*mxNC5 (%)*	*mxC4 (%)*	*mxNC4 (%)*	*mdC5 (%)*	*mdNC5 (%)*	*mdC4 (%)*	*mdNC4 (%)*
Present	95 (81.9)	96 (82.8)	108 (93.1)	105 (90.5)	108 (93.1)	109 (94.0)	106 (91.4)	106 (91.4)
Agenesis	14 (12.1)	12 (10.3)	0 (0.0)	0 (0.0)	6 (5.2)	5 (4.3)	0 (0.0)	1 (0.9)
Extracted for treatment	7 (6.0)	8 (6.9)	8 (6.9)	11 (9.5)	2 (1.7)	2 (1.7)	10 (8.6)	9 (7.8)

* mx = maxillary; md = mandibular; C = cleft side; NC = noncleft side; 5 = second premolar; 4 = first premolar.

TABLE 6 Orthognathic Surgery

	No. of Patients (% of Total)
No orthognathic surgery	40 (34.5)
Orthognathic surgery with segmental advancement (two-piece Le Fort I)	40 (34.5)
Orthognathic surgery without segmental advancement (one-piece Le Fort I)	36 (31.0)

Discussion

The present study identified the dental irregularities associated with CUCLP and their clinical management at SickKids Hospital. Of particular interest were the maxillary permanent lateral incisors and the premolars. Orthodontic and surgical treatment often must compensate for the three most commonly encountered problems: agenesis of lateral incisors, the alveolar cleft, and a deficient maxilla.

Only 7% of the cases reviewed had a permanent lateral incisor on the cleft side at the end of treatment. This is in agreement with the findings of Tan et al. (1996), who also reported that 7% of the cases from Princess Margaret Hospital for Children in Perth, Australia, finished with a lateral incisor in place.

The agenesis of permanent teeth is significantly higher in children with CUCLP, both in and outside the cleft region. A review of the literature (Ranta, 1986) showed that agenesis of teeth outside the cleft site occurs in descending frequency from the maxillary second premolar (7.5% to 32.3%) to the permanent maxillary lateral incisor on the noncleft side (3.1% to 10.4%) to the mandibular second premolar (0.4% to 10.8%). The current study found agenesis of 11.2% of the maxillary second premolars, 12.0% of the noncleft-side maxillary lateral incisors, and 3.4% of the mandibular second premolars. These numbers are mostly higher than the frequency of agenesis in the normal population: 2.2% for the maxillary lateral incisor and 3.4% to 6.6% for the mandibular and maxillary second premolars, respectively (Symons et al., 1993).

On reviewing the literature, it is difficult to compare studies based on the incidence of hypodontia in patients with cleft lip and/or cleft palate and CUCLP, particularly of the lateral incisor on the cleft side. These difficulties are due to the wide variety of methods that researchers use to report their data: number of laterals missing in the sample; number of subjects with missing laterals; frequency of missing laterals in the total number of collected teeth; distinguishing between true lateral incisors (based on position or size and shape); and supernumeraries or grouping them in the same category. To further complicate comparisons among studies, various cleft types and severities are examined together when analyzing data within the same study. In this study, only subjects with history of CUCLP were considered.

Shapira et al. (2000) found agenesis of the mesial cleft-side maxillary permanent lateral incisor to occur in 74.0% of the cases in their sample, which is consistent with the 75.9% found in the present study. Böhn (1963) found the same in 45.9% of the cases; Hellquist et al. (1979) found the same in 42.6% of the cases; and Dixon (1966) found the same in 39.0% of the cases (Table 9). These differences are due largely to the variable cleft types and severities included in each of the above studies.

Many studies have shown that a conical pattern of the cleft-side lateral incisor is more frequent than a shape similar to the antimere (Böhn, 1950; Nagai et al., 1965; Ranta, 1972a; Vichi and Franchi, 1995; Tsai et al., 1998). In the current study, only 3.6% of cases had a Lm that was normal in crown and root morphology at treatment completion. This is similar to a study by Hellquist et al. (1979) that found 6.9% of 172 cases to have a normal (i.e., not malformed) permanent maxillary lateral incisor on the cleft side. Hellquist and colleagues' study, however, included patients with a unilateral cleft of the lip and alveolar process and patients with CUCLP.

Table 10 compares the percentages of agenesis of teeth outside the cleft area found in the present study with those in the previously published literature.

TABLE 7 Prosthodontic Management

	No. of Patients (% of Total)
Not required*	93 (80.2)
Fixed prosthesis (bridge)	2 (1.7)
Removable prosthesis	6 (5.2)
Implant	12 (10.3)
No prosthesis (failed cases)†	3 (2.6)

* Not required due to presence of lateral incisor or space closure with orthodontic treatment following bone graft with or without orthognathic surgery.

† Failed cases due to patients not proceeding with recommended prosthetic treatment for the cleft-side lateral incisor.

TABLE 8 Comparison of Treatment Rendered Between Two Time Periods (n = 116)

	1990s	*2000s*		
	n = 42	*n = 74*	*Difference*	*p Value*
Patients receiving segmental surgery (%)	28.6	37.8	+9.2	$p \leq 1.0$
Patients receiving implant (%)	7.1	12.2	+5.1	$p \leq 1.0$
Patients receiving fixed bridge (%)	4.8	0.0	−4.8	$p \leq .10$
Patients with lateral incisor at treatment finish (%)	7.1	6.8	−0.3	$p \leq 1.0$

TABLE 9 Comparison of Percentages of Missing Cleft-side Maxillary Lateral Incisors

Study	*Agenesis of the Cleft-side Maxillary Lateral Incisor (%)*	*No. of Subjects in the Study*	*Types of Clefts Included*
Present study	75.9	116	CUCLP only*
Shapira et al. (2000)	74.0	278	Cleft lip and/or palate
Böhn (1963)	45.9	281	Cleft lip and/or palate
Hellquist et al. (1979)	42.6	172	Unilateral cleft lip and alveolus and CUCLP
Dixon (1966)	39.0	100	Cleft lip and/or palate

* CUCLP = complete unilateral cleft lip and palate.

TABLE 10 Comparison of Percentages of Missing Teeth Outside the Cleft Area in Three Studies

Study	*Present Study*	*Ranta (1986)*	*Hellquist et al. (1979)*
Maxillary lateral incisor on noncleft side	12.1	10.4	3.1
Maxillary second premolar	10.3 on NC side* 12.1 on C side	32.3	7.5
Mandibular second premolar	4.3 on NC side* 5.2 on C side	7.6	0.4
Other teeth		1.1	0.1

* The present study was the only study cited above that distinguished between agenesis of second premolars on the cleft (C) versus the noncleft (NC) side.

The second tooth type most frequently missing according to the literature is the maxillary second premolar. The present study found that the cleft-side maxillary second premolars were the most frequently missing premolars, with a frequency of 12.0%, compared with a frequency of noncleft-side maxillary second premolar agenesis of 10.3%. These results were comparable to previous study results. Shapira et al. (1999), in their sample of various cleft types, found the second premolars to be missing in 18% of the cases. They also found that when present, the second premolars typically showed a delay in development and timing of eruption, which was thought to represent a mild expression of hypodontia (Shapira et al., 1999).

Shapira et al. (2000) found that the cleft-side second premolar was missing approximately twice as often in the maxilla as in the mandible. This finding was confirmed in our material. Other studies have shown that hypodontia of second premolars is most common on the maxillary cleft side (Olin, 1964; Hellquist et al., 1979).

The maxillary noncleft second premolar was missing approximately twice as often (10.3%) as the mandibular noncleft-side second premolar (average 4.3%). Shapira et al. (2000) reported smaller percentages of 3.0% and 1.5%, respectively. It is evident in the literature that most studies have shown there is no clear difference in frequency of agenesis between the cleft and noncleft sides of the mandible.

The findings of this study reflect the clinical judgment of the treating practitioners and are not intended as justification for extracting or maintaining specific teeth. Most of these practitioners showed a preference for extracting small laterals on the cleft side that had compromised bone levels before ABG and orthodontic treatment. Trying to save these teeth increases the difficulty of prosthetic treatment. More importantly, it increases the burden of treatment on already overburdened patients. Both Lm and Ld are helpful in maintaining surrounding bone and therefore, must be preserved until an ABG is performed. Extracting the abnormal lateral incisor around the time of alveolar bone grafting allows the canines to migrate and erupt forward through the grafted area, providing an improved bony environment that facilitates orthodontic and prosthodontic treatment and improves the stability and health of the periodontium (El Deeb, 1982; Hall and Posnick, 1983; Hinrichs et al., 1984). Canine substitution is often a superior alternative to these compromised lateral incisors due to better bone support, larger root structure, and improved aesthetics.

Patients with cleft lip and palate have a markedly higher frequency of agenesis of permanent teeth than the general population has. Many theories have been advanced attempting to explain this finding. These theories include multiple genetic and environmental factors, mesenchyme deficiency, and direct effect of the cleft on the primordial tissues related to the development of the lateral incisor (Ross and Johnston, 1972). It is interesting to note, however, that no association was found between missing maxillary second premolars and missing cleft-side lateral incisors. According to the earlier protocol at SickKids, when a patient was deemed to require a Le Fort I maxillary advancement in the future, bone grafting often was delayed until that time to avoid a second surgery. Contrary to this, in the late 1990s a shift occurred in the protocol, which called for mixed-dentition bone grafting even for patients deemed to require eventual Le Fort I surgery at skeletal maturity. This was a result of internal audits showing better bone support and periodontal status in the patients who received earlier bone grafts. Bone grafting is commonly done prior to eruption of the canine to allow the canine to erupt through the grafted bone. This helps to consolidate the graft and encourages the canine to erupt mesially, close to the central incisor in cases where the lateral incisor is absent, thus facilitating orthodontic space closure. Distalization of a mesially erupted canine in patients where an implant is deemed appropriate allows future implant site development by building up the alveolar ridge through orthodontic tooth movement (Kokich, 2004).

Boyne and Sands (1976) proposed that when bone grafts are delayed until the late mixed-dentition stage, there are periodontal and restorative challenges due to significant inadequacies of bone at the cleft alveolar site. For this reason, some institutions advocate bone grafting of the alveolar cleft during the late primary or early mixed-dentition stage, at about 5.5 to 6.5 years, depending on dental development (Precious et al., 2001). There is some concern, however, that maxillary growth may be interfered with to some extent in this approach.

The two main methods to restore the alveolar cleft in the absence of the lateral incisor are dental space closure (cases finishing with the canine beside the central incisor, performed in 80% of cases in the present study) and dental space opening (performed in 17% of cases in this study, 10% of cases using a single tooth implant, and 7% of cases using a removable prosthesis). Of the 80% of subjects who had the space closed, 45% had orthodontic closure and 35% had surgical advancement of the posterior segments. Reducing the number of lateral incisor prostheses (single tooth implants, bridgework, removable prostheses) and increasing the number of canine-lateral substitutions were considered more desirable, when possible, because they

reduce the lifetime maintenance and cost for the patient. Enemark et al. (1985) reported a closed dental arch in 40% of their patients with CL/P, without a cleft-side lateral incisor, using conventional intraoral orthodontic appliances after alveolar bone grafting. They reported that a closed dental arch could have been achieved in a higher percentage of patients; however, aesthetically, they believed that facial appearance is often improved by sagittal expansion of the maxillary teeth, thereby opening up for prosthodontic bridgework in the cleft area.

It was interesting to compare the variation in treatment protocols within the two time periods studied (Table 8). There were only minor differences between the two time periods, indicating that treatment protocols stayed relatively similar, perhaps because the outcomes continued to be considered successful.

Ten cases of transposition of the maxillary canine and first premolar were found in this study (8.6% of cases). No other studies on transpositions have been reported in the cleft population. When compared with the literature in the general population, however, this rate was highly significant ($p < .0001$). Various studies that recorded transpositions of the same teeth were of much lower prevalence in the noncleft population: 0.03% in Swedish school children (Thilander and Jakobsson, 1968), 0.13% in Saudi Arabian dental patients (Ruprecht et al., 1985), and 0.25% in Scottish orthodontic patients (Sandham and Harvie, 1985).

Conclusions

In patients with CUCLP from our sample:

- Cleft-side laterals were rarely present at the conclusion of treatment (7%).
- When cleft-side lateral incisors developed they were frequently abnormal in size, shape, and bone support, and they were commonly extracted in favor of canine substitution.
- The cleft-side lateral incisor space was closed by orthodontics or by surgery with canine substitution in 80% of cases.
- Prosthetic replacement of the cleft-side lateral incisor was performed in 20% of cases.
- Agenesis of the cleft-side second premolar was not correlated with the agenesis of the cleft-side lateral incisor.
- There was a statistically significantly higher number of transpositions of maxillary canines and first premolars (8.6% of cases) when compared with the general population.

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