

Dental anomalies in children with cleft lip and palate in Western Australia

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ABSTRACT

Objective: The purpose of this paper was to describe the prevalence and type of dental anomalies in the primary and permanent dentition in children with a cleft condition at Princess Margaret Hospital in Perth, Western Australia. **Materials and Methods:** The details of 162 current dental patients extracted from the main dental database through their year of birth for the period 1998–2001 were selected consecutively. Dental records and X-rays were examined by one examiner (WN) and verified by a second examiner (RB) to determine dental development. The mean age of the subjects was 10.8 years with equal numbers of males and females. Subjects were further divided into cleft type; unilateral cleft lip (UCL) and palate, bilateral cleft lip (BCL) and palate, UCL, BCL, and cleft palate. **Results:** One hundred sixty-two subjects were grouped into 21 categories of anomaly or abnormality. Prevalence rates for the categories were calculated for the overall group and for gender and cleft type. **Conclusion:** Overall, 94% of patients were found to have at least one dental anomaly, with fifty-six (34%) patients having more than one anomaly or abnormality.

Key words: Cleft lip and palate, dental anomalies, Western Australia

INTRODUCTION

The occurrence of dental anomalies may result from both environmental and genetic factors. Previous studies indicate that there is a higher prevalence of dental anomalies in children with a cleft condition than in the general population.^[1,2] Studies have also demonstrated that both genetics and the surgical repair of the palate influence the occurrence of dental anomalies in the cleft population.^[3] Anomalies include variations in tooth size, shape, number, structure and formation, and eruption timing.^[2]

Both sets of dentition may be affected and occur more frequently on the cleft-affected side of the maxilla.^[4] The most common types of dental anomalies have been reported to be absent or supernumerary teeth,

enamel dysplasia and discoloration and delayed root development. In children with cleft lip and palate, the lateral incisor in the alveolar cleft region has the highest prevalence of dental developmental disorders.^[2] This may cause functional and esthetic issues for the child and complicating factors for dental and orthodontic treatment.

This paper will report the prevalence and type of dental anomalies in the primary and permanent dentition in children with a cleft condition at Princess Margaret Hospital in Perth, Western Australia.

Subjects

The details of current dental patients were selected consecutively from the main dental database via their

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year of birth for the period 1998–2001. Patients who had not attended for the past 3 years, those with a syndrome and patients without a cleft condition were excluded, leaving a total of 172 subjects for this study. Ten subjects did not have any dental anomalies and were excluded, leaving 162 subjects with at least one anomaly. The majority of subjects (92%) were Caucasian with 84% residing in the Metropolitan area. The mean age of the subjects was 10.8 years (age range of 8.9–11.9 years) with equal numbers of males and females.

MATERIALS AND METHODS

Dental records and X-rays were examined by one examiner (WN) and verified by a second examiner (RB) to determine dental development. Subjects were further divided into cleft type; unilateral cleft lip and palate (UCLP: 25% [left 14%; right 11%]), bilateral cleft lip and palate (BCLP: 8.6%), UCL: 20%, BCL: 1.2%, and isolated cleft palate (CP: 45%). The following anomalies were reported: Agenesis, crowding, delayed development, demineralization, dysplastic, early loss, ectopic, fissural, hypomineralization, hypoplastic, microform, peg lateral, pits and fissures, premaxilla, resorptive, retained, rotated, supernumerary, transposition, miscellaneous dental anomalies, nil and other [Appendix 1 for definitions].

Statistical analysis

The rates of occurrence of each anomaly were calculated as a percentage of the total sample in each group. Microsoft Excel 2016 (Microsoft Corporation, One Microsoft Way Redmond, WA 98052-7329, USA) was used for data storage, calculating descriptive statistics, and chart generation. Incidence rate difference was analyzed with Wilcoxon statistical test using Minitab® version 17 (Minitab Inc., Quality Plaza, 1829 Pine Hall Road, State College, PA 16801-3210, USA). Chi-square analysis was used to determine if any significant difference existed between genders.

RESULTS

One hundred sixty-two subjects were grouped into 21 categories of anomaly or abnormality [Appendix 1 for definitions]. Prevalence rates for the categories were calculated for the complete group. Overall, 94% of patients were found to have at least one dental anomaly [Figure 1] with 56 (34%); patients having more than one anomaly or abnormality (65% had one reported anomaly; 22% had two; 11% had three; 1.2% had four, and 0.6% had five reported anomalies). Dental anomalies and abnormalities were also calculated by

gender and cleft type [Tables 1 and 2]. A significant statistical difference was not found between the frequency of dental anomalies and gender ($P < 0.01$).

Agenesis was the most prevalent anomaly in this study (15%) [Table 1]. The second most frequently occurring abnormality was crowding. This affected 14% (22) of patients, predominantly female, and with a CP condition. Supernumerary teeth were found to be the third most commonly occurring dental anomaly affecting 10% (16) of patients. The most frequently affected tooth location was 62.

With the exception of demineralization and ectopic eruption, all dental anomalies occurred more frequently on the left side. Seventy-five percent of agenesis occurred more often on the left side than on the right side. Dental anomalies affecting tooth shape occurred exclusively on the cleft-affected side in this study. The most frequently occurring shape anomaly was the occurrence of a peg or conical shaped lateral incisor, and this most commonly (95%) affected tooth 22.

Fourteen percent (23) of patients had anomalies that occurred as a single condition or very low numbers. These were categorized as miscellaneous dental anomalies and was comprised crib tooth, star lateral, fusion, cavitation, severe enamel staining, and submerging teeth (2), cross-bite, hypomineralization, and palatal eruption (4), transverse migration, deep retentive fissures, necrotic pulp space, and distal flaring.

Six percent (10) of patients had anomalies that were categorized as “other” and was comprised cystic hemangioma, palatal constriction, gingival hyperplasia, limited opening, polyps, abnormal frenum, and glossitis.

Absent teeth (due to agenesis) were equally represented (50%) by gender [Table 1]. Crowding (82%) resorptive (88%) and retained teeth (85%) were more prevalent in females in this study. Anomalies of fissural teeth (58%), supernumerary (63%), and rotated teeth (60%) were more prevalent in males.

When dental anomalies were compared by cleft type [Table 2] agenesis occurred most frequently in patients with a cleft of the lip and palate (50%). Other anomalies that were more prevalent in this group were fissural (68%), hypoplastic (60%), and transpositioned (100%) teeth. Crowding (73%), demineralization (56%), resorptive (75%), and miscellaneous (61%) anomalies occurred more frequently in patients

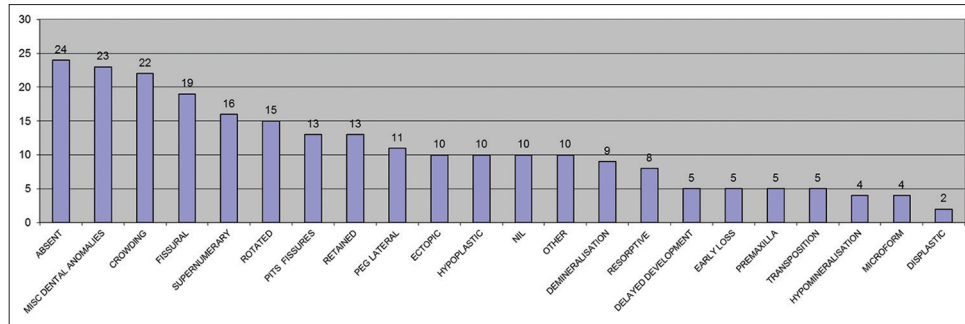


Figure 1: Occurrence of dental anomalies for all patients

Anomaly	All patients (percentage of subjects)	Percentage of anomaly	
		Female	Male
Agenesis	24 (15)	12 (50)	12 (50)
Crowding	22 (14)	18 (82)	4 (18)
Delayed development	5 (3)	1 (20)	4 (80)
Demineralization	9 (6)	4 (44)	5 (56)
Dysplastic	2 (1)	0 (0)	2 (100)
Early loss	5 (3)	1 (20)	4 (80)
Ectopic	10 (6)	6 (60)	4 (40)
Fissural	19 (12)	8 (42)	11 (58)
Hypomineralization	4 (2)	1 (25)	3 (75)
Hypoplastic	10 (6)	3 (30)	7 (70)
Microform	4 (2)	3 (75)	1 (25)
Peg lateral	11 (7)	7 (64)	4 (36)
Pits and fissures	13 (8)	7 (54)	6 (46)
Premaxilla	5 (3)	1 (20)	4 (80)
Resorptive	8 (5)	7 (88)	1 (13)
Retained	13 (8)	11 (85)	2 (15)
Rotated	15 (9)	6 (40)	9 (60)
Supernumerary	16 (10)	6 (38)	10 (63)
Transposition	5 (3)	3 (60)	2 (40)
Misc dental anomalies	23 (14)	12 (52)	11 (48)
Nil	10 (6)	5 (50)	5 (50)
Other	10 (6)	8 (80)	2 (20)

with a CP. Supernumerary teeth (63%) occurred more frequently in patients with a cleft lip only. When compared with further defined cleft types [Table 3], the most frequent anomaly per cleft type are as follows; tooth shape disorder of peg lateral (9%) occurred most frequently in BCL; absent (16%) and premaxilla abnormality (16%) in BCLP; crowding (72%) and other miscellaneous dental anomalies (60%) in CP; supernumerary (62%) and fissural and rotated teeth (both 31%) in UCL; absent (25%) and fissural (26%) in ULCLP and fissural (26%) in URCLP.

The 10 patients whom were found to have no anomalies were represented by equal numbers of male to female and classified within the following cleft types; six UCLP patients, three CP patients, and one UCL patient.

DISCUSSION

A higher prevalence of dental anomalies has been reported in cleft populations compared with the general population^[1-10] and in Caucasian subjects.^[9] Anomalies reported include variance in tooth shape, size, color, structure, position, and the influence of the area of the repaired cleft defect. This study investigated the prevalence of dental anomalies and abnormalities in a predominantly Caucasian cleft population.

Agenesis was the most prevalent anomaly in this study, supporting results reported in other studies of 50% and greater.^[3,4] In this study, the rates of agenesis on the cleft-affected side varied between 75% and 92%, depending on the type of cleft type, with statistically significant differences for CP ($P < 0.05$), and cleft lip and palate ($P < 0.01$). Previous studies have reported agenesis of the permanent lateral incisor on the cleft-related side to be the most frequently occurring anomaly in children with a cleft condition.^[1-6] This study also found that the permanent maxillary lateral incisor in the area of the cleft was the tooth most frequently absent (64%). Lourenço Ribeiro *et al.*^[1] suggest that this may be due to the compromised blood supply in the cleft-affected area, either as a congenital condition or the result of surgical repair.

Supernumerary teeth have been reported to be the second most frequently occurring dental anomaly in the literature.^[1,2] Akcam *et al.*^[3] report the prevalence of supernumerary to be from 1.9% to 10.0% in the UCLP and CP groups. They further reported a higher rate (22%) of supernumerary teeth present in the permanent dentition in the cleft area in subjects with UCLP or CP. It has been suggested that supernumerary teeth in cleft lip and palate result during cleft formation from fragmented lamina of the dentition. In this study, supernumerary teeth were found to be the third most frequently occurring dental anomaly. The most commonly affected site was at tooth location 62.

Table 2: Dental anomalies by broad cleft type

Anomaly	Lip (%)	Palate (%)	Lip and palate (%)	Total
Agenesis	3 (13)	9 (38)	12 (50)	24
Crowding	2 (9)	16 (73)	4 (18)	22
Delayed development	2 (40)	2 (40)	1 (20)	5
Demineralization	1 (11)	5 (56)	3 (33)	9
Dysplastic	0 (0)	1 (50)	1 (50)	2
Early loss	1 (20)	4 (80)	0 (0)	5
Ectopic	1 (10)	4 (40)	5 (50)	10
Fissural	6 (32)	0 (0)	13 (68)	19
Hypomineralization	1 (25)	2 (50)	1 (25)	4
Hypoplastic	3 (30)	1 (10)	6 (60)	10
Microform	1 (25)	0 (0)	3 (75)	4
Peg lateral	4 (36)	4 (36)	3 (27)	11
Pits fissures	1 (8)	8 (62)	4 (31)	13
Premaxilla	0 (0)	0 (0)	5 (100)	5
Resorptive	0 (0)	6 (75)	2 (25)	8
Retained	0 (0)	6 (46)	7 (54)	13
Rotated	6 (40)	4 (27)	5 (33)	15
Supernumerary	10 (63)	1 (6)	5 (31)	16
Transposition	0 (0)	0 (0)	5 (100)	5
Misc dental anomalies	3 (13)	14 (61)	6 (26)	23
Nil	6 (60)	4 (40)	0 (0)	10
Other	0 (0)	7 (70)	3 (30)	10

Table 3: Dental anomalies by refined cleft type

Anomaly	BCL	BCLP	CP	UCL	ULCLP	URCLP	Total
Absent	0	4	9	3	6	2	24
Crowding	0	1	16	2	2	1	22
Delayed development	0	0	2	2	1	0	5
Demineralization	0	0	5	1	0	3	9
Dysplastic	0	0	1	0	1	0	2
Early lo	0	0	4	1	0	0	5
Ectopic	0	0	4	1	2	3	10
Fissural	0	3	0	6	5	5	19
Hypomineralization	0	0	2	1	1	0	4
Hypoplastic	0	2	1	3	3	1	10
Microform	0	1	0	1	1	1	4
Peg lateral	1	0	4	3	2	1	11
Pits fissures	0	0	8	1	3	1	13
Premaxilla	0	4	0	0	1	0	5
Resorptive	0	1	6	0	1	0	8
Retained	0	2	6	0	2	3	13
Rotated	0	1	4	6	2	2	15
Supernumerary	0	1	1	10	2	2	16
Transposition	0	2	0	0	1	2	5
Misc dental anom	1	1	14	2	4	1	23
Nil	0	0	4	6	0	0	10
Other	0	0	7	0	1	2	10

BCL: Bilateral cleft lip, BCLP: Bilateral cleft lip and palate, CP: Cleft palate, UCL: Unilateral cleft lip, ULCLP: Unilateral cleft lip and palate, URCLP: Unilateral right cleft lip and palate

In UCLP patients, it has been reported that dental anomalies occur more frequently on the left side (ratio of 2:1).^[2,3] The results from this study supported

those findings overall, with the majority of dental anomalies across all cleft types, occurring more frequently on the left side including 75% of agenesis. However, this study also found some differences; agenesis having a 3:1 ratio, miscellaneous anomalies having a ratio of 4:1, and demineralization occurred exclusively on the right side.

Dental anomalies affecting tooth shape occurred exclusively on the cleft-affected side in this study. The most frequently occurring tooth shape anomaly was the occurrence of a peg or conical shaped lateral incisor, and this most commonly (95%) affected tooth 22. Previous research has shown the prevalence of microdontia to vary in the general population from 1.5% to 2.0%.^[3] In this study, when comparing to the general population, there was a only a slightly higher prevalence of microdontia (1.9–2.4%) on the cleft side for both the UCLP groups. Tooth anomalies in anterior regions are the most obvious and not only influence appearance but may prove a challenge due to compromised root formation and positioning to achieve the best esthetic outcome. Satisfaction with dental esthetic outcome may be very important to a child with a cleft condition that already has a visible difference in appearance from facial asymmetry or scarring from cleft-related surgical repair.

It has been reported that in UCLP patients, dental anomalies occur with a higher frequency on the cleft-affected side.^[3] This study supported the higher occurrence of anomalies on the cleft-affected side with 95% of dental anomalies occurring on the side of the cleft.^[4-7] This occurrence has been suggested to be due to etiological factors involved with a cleft formation, which also influence dental formation.

Trotman *et al.*^[8] suggested that the forms of dental anomalies and abnormalities may be related to different cleft types, primarily influenced by the developmental effect of clefting process, and subsequent cleft repair. They further suggest that there may an influential link between the genetic factors, dental anomalies, and cleft type. This study found agenesis to be the most frequent in clefts of the lip and palate, crowding in isolated CP, and the presence of supernumerary teeth in isolated cleft lip.

CONCLUSION

A very high proportion of subjects in this study were found to have at least one dental anomaly, with agenesis being found to be the most frequently

reported anomaly occurring equally between genders and most frequently in patients with a cleft of the lip and palate. Dental anomalies in patients with a cleft condition may be a complicating factor and must be taken into consideration as part of the treatment planning process and should be undertaken by, or in consultation with, pediatric dental specialists to achieve the best esthetic and functional outcome.

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Conflicts of interest

There are no conflicts of interest.

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APPENDIX

Appendix 1: Anomaly definitions

1. Agenesis: Congenital absence of a permanent tooth or germ
2. Crowding: Discrepancy between tooth size and jaw size
3. Delayed development: Delayed tooth eruption for developmental timing
4. Demineralization: Loss of tooth enamel
5. Dysplastic: Normal tooth enamel with atypical dentin and abnormal pulp morphology
6. Early loss: Early exfoliation for developmental age
7. Ectopic: Eruption of teeth outside of the usual developmental course, malposition of permanent tooth bud
8. Fissural: Development of a tooth in the area of the cleft palate repair
9. Hypomineralization: Tooth enamel defect
10. Hypoplastic: Where the dental enamel shows surface loss or a break in continuity
11. Microform: One or more teeth appear smaller than surrounding teeth
12. Peg lateral: Conical-shaped lateral tooth
13. Pits and fissures: Imperfection of the enamel
14. Premaxilla: Abnormal position or development of the premaxilla
15. Resorptive: Loss of tooth root structure, dentin, or bone
16. Retained: Delayed eruption of the permanent teeth by approximately 1 year of normal developmental timing
17. Rotated: Mesio or disto-lingual intra-alveolar displacement of teeth
18. Supernumerary teeth: Those which are additional to the regular number of teeth
19. Transposition: Teeth are juxta-positioned with the neighboring teeth
20. Miscellaneous dental anomalies: Single anomaly of: Crib tooth, star lateral, fusion, cavitation, severe enamel staining, submerging teeth, cross-bite, hypomineralization, palatal eruption, transverse migration, deep retentive fissures, necrotic pulp space, and distal flaring
21. Nil: No abnormality present
22. Other: Single anomaly of: Cystic hemangioma, palatal constriction, gingival hyperplasia, limited opening, polyps, abnormal frenum, and glossitis