The Oral Health of Children With Clefts of the Lip, Palate, or Both

J.R. CHAPPLE, B.D.S., M.F.D.S.
J.H. NUNN, PH.D., F.D.S., D.D.P.H., B.D.S.

Objective: **The purpose of this study was to assess the prevalence of dental caries, developmental defects of enamel, and related factors in children with clefts.**

Design: **This cross-sectional prevalence study used standard dental indices for assessment.**

Setting: **Children underwent a dental examination under standard conditions of seating and lighting in the outpatient department of a dental hospital as part of an ongoing audit to monitor clinical outcomes.**

Participants: **Ninety-one children aged 4, 8, and 12 years were included in the study.**

Outcome Measurements **Dental caries were assessed by use of the decayed, missing, and filled index for primary teeth (dmft); Decayed, Missing, and Filled index for permanent teeth (DMFT) according to the criteria as used in the national survey of children's dental health in the United Kingdom (O'Brien, 1994). Developmental defects were assessed using the modified Developmental Defects of Enamel Index (Clarkson and O'Mullane, 1989). Dental erosion was assessed using the criteria derived for the national survey of children's dental health (O'Brien, 1994).**

Results: **Caries prevalence increased with age; 63% of patients at 4 years and 34% at 12 years were caries free. The mean dmft for the 4-year-olds was 1.3 with a mean DMFT for the 12-year-olds of 1.8. All the 4-year-olds had evidence of erosion of enamel in the primary teeth (incisors and first molars) and 56% of the 12-year-olds had erosion of permanent teeth (incisors and first permanent molars). Developmental defects of enamel became more prevalent with age, with at least one opacity in 56% of 4-year-olds and 100% of 12-year-olds. Hypoplasia was not found in the primary dentition but affected permanent teeth in 38% of 8-year-olds and 23% of the 12-year-olds.**

Conclusion: **This study has shown that dental disease is prevalent in these patients. These assessments not only provide a baseline on oral health parameters in young people with clefts but underline the need for a more aggressive approach to prevention of oral disease to optimize clinical outcome.**

KEY WORDS: *clefts, dental caries, dental health, fluoride, hypoplasia*

Patients who have clefts of the lip and palate often need complex and extensive treatment, usually provided by an interdisciplinary team of orthodontists, plastic surgeons, pediatric dentists, maxillofacial surgeons, speech therapists, and others.

Following surgical repair of the cleft, the long-term oral health management of patients centers around dental caries, malocclusions, hypoplasias, and gingivitis. In some cases, the prevalence of these conditions is greater in children with clefts as compared with their peers. Bokhout et al. (1997) published results of a survey of 81 preschool children with clefts and a control group of 77 children with no congenital malformation. Sixty-nine percent of the children with clefts were caries free in the primary dentition, compared with 93% of the control children.

A year later, Paul and Brandt (1998) published results of a study of children with clefts in the south of England. The authors examined 114 children aged 3 to 18 years of age. Looking at both dentitions combined, 54% of the children were caries free. This study highlighted differences between different ethnic groups such that children from the Indian subcontinent had a higher mean caries experience than did Caucasian children.

More recently published data on the oral health of children

Mr. Chapple is a General Dental Practitioner, Dental Hospital, Newcastle upon Tyne, United Kingdom. Dr. Nunn is Senior Lecturer/Honorary Consultant, School of Dentistry, Newcastle upon Tyne, United Kingdom.

Presented at the Biennial Congress of the International Association of Paediatric Dentistry, Queen Elizabeth II Conference Centre, September 2–4 1999; London, England.

Submitted February 2000; Accepted October 2000.

Reprint requests: Dr. June Nunn, Department of Child Dental Health, School of Dentistry, Framlington Place, Newcastle upon Tyne NE2 4BW, United Kingdom. E-mail j.h.nunn@ncl.ac.uk.

with cleft lip and palate comes from Northern Ireland. Gregg et al. (1999) published results to the Irish division of the International Association for Dental Research from data collected at joint outpatient cleft lip and palate clinics. Their findings showed that 50% of the 5-year-olds and 40% of the 12-year-olds were caries free. A related finding was that caries experience was inversely proportional to fluoride supplement uptake.

A similar study was recently undertaken in the Bristol Dental Hospital, England (Rivkin et al., 1999). This study looked at attendance and caries experience of 85 children with cleft lip and palate. The majority of the children with clefts (41%) had isolated clefts of the palate only. The results indicated that 51% of 5-year-olds and 54% of the 10-year-olds were caries free in the primary dentition and 73% were caries free in the permanent dentition; 50% of 15-year-olds had a caries-free permanent dentition, but none of the 20-year-olds were caries free. The majority of children and young people claimed to be seeing a dentist regularly (92%) with 76%, 2%, and 15%, respectively, using the general, community, and hospital dental services.

A variety of reasons has been postulated for the high caries prevalence in this group of patients. One reason may be the irregularity of teeth (Wong and King, 1998), and it is one rationale for aligning teeth in these young people. Another reason may be the paucity of advice that the parents get on feeding and oral hygiene. Fitzgerald et al. (1990) showed that the majority of advice received by parents was given by a voluntary parent's organization only.

A number of researchers have published results of studies, both clinical (Olin, 1964; Dixon, 1968; Ranta, 1986; Vichi and Franchi, 1995; Malanczuk et al., 1999) and histological (Jordan et al., 1966; Lagarde et al., 1987, 1989), on the prevalence of enamel defects in children with clefts. The precise etiology of these enamel defects remains unclear.

The purpose of this paper is to look at the relationship among caries prevalence, fluoride uptake, and enamel hypoplasia in child patients with a cleft living in a partly fluoridated region in northeast England.

Material and Method

Subjects for the study were recruited from monthly cleft audit clinics run within the outpatient department of the Newcastle Dental Hospital. Families of children and young people aged 4, 8, and 12 years of age at the time of the audit were invited by letter to attend the clinic. The letter described the purpose of the clinic and what would be required of the family in terms of time spent in the department.

On the examination day, children and one or both parents were seen first by the plastic surgeon for a structured interview covering areas of patient and parent satisfaction with both the process and outcome of clinical care to date. A speech assessment, including video-recordings was made, and standard clinical photographs were taken. A full dental examination was carried out to include assessments for caries, dental erosion, and developmental defects of enamel. An orthodontic assessment included study model impressions and appropriate radiographs were undertaken.

TABLE 1 Patient Details

Age	4 y	8 y	12 y
N =	19	28	44
Cleft type	lip	palate	lip and palate
N =	24	41	26
Fluoride level	full	part	none
N =	50	16	25

The criteria for the dental examinations were as follows: for dental caries and erosion, the criteria were the same as those used in the children's dental health survey (O'Brien, 1994). The criteria for assessment of developmental defects of enamel (enamel opacities and hypoplasia) were the modified Developmental Defects of Enamel index (Clarkson and O'Mullane, 1989). At the same time as the clinical examination, the child or the parent was asked if the family had a dentist that the child attended and questions were asked also to assess the child's exposure to systemic fluorides: either from a fluoridated water supply or from the long-term use of supplements. A note was made of the child's cleft type.

The clinical examination was undertaken with the child supine in a dental chair and using an operating light, mouth mirror, and probe, the latter for the purpose of clearing away debris on a surface which was otherwise obscured. The teeth were examined dry for the purposes of assessing dental caries and erosion. Bitewing radiographs were taken if the child had no current views available.

Results

Table 1 describes the sample, including their exposure to systemic fluorides available during the period of dental development. The majority of children seen were in the 12-year-old group. The distribution of cleft type was similar to the normal distribution for cleft patients, with predominantly more children having isolated clefts of the palate. Of the children attending the audit clinics, most had full exposure to some form of systemic fluorides full time (N = 50), while 25 had no such use.

Data for dental caries for boys and girls have been combined because there was no significant difference between the groups. Overall, 41% of the children were caries free in one or both dentitions. The mean dental caries experience in both the primary and permanent dentition increased with increasing age (Table 2). Although nearly two-thirds of 4-year-olds were free of decay experience, just over one-third of the 12-year-olds were caries free.

Developmental defects of enamel primarily record colored areas on the surface of the enamel as well as hypoplastic areas (i.e., areas in which the enamel is missing to a greater or lesser extent). Enamel opacities as recorded in this study were minimal, and data for these have thus not been presented here. Of

TABLE 2 Dental Caries Experience in the Primary and Permanent Dentition For the Three Age Groups (Boys and Girls Combined)*

	Age, y		
Variable	*4*	*8*	*12*
dmft (mean)	1.3	1.8	0.9
95% CI	0.2–2.4	0.9–2.8	0.4–1.4
DMFT (mean)		0.4	1.8
95% CI		0.1–0.7	0.9–2.3
Caries free (%)	63	36	34

* dmft = decayed, missing, and filled index for primary teeth; DMFT = Decayed, Missing, and Filled index for permanent teeth.

more interest in children with clefts was the prevalence of enamel hypoplasia, especially on teeth developing in the area of the cleft.

Enamel hypoplasia was prevalent in this group of children (24%). The analysis was undertaken according to cleft type and whether the hypoplasia presented on either the anterior or posterior teeth (Table 3). Children with clefts involving the anterior part of the mouth were more likely to have hypoplasia of the anterior teeth, although the numbers are small so that this indicates a trend only.

Fluoride status was assessed according to the child's lifetime history of exposure to systemic forms of fluoride (water or supplements). Full-time exposure appeared to confer benefits; children who had had access to either fluoridated water or fluoride supplements had a lower mean caries experience than did children with no systemic fluoride ingestion history (Table 4). ''Part-time'' status reflected the histories of those children who had either moved in or out of a fluoridated water area or who had taken fluoride supplements for only part of their life.

The cleft type did not appear to significantly affect a child's predisposition to dental caries in the permanent dentition with no discernible trend emerging between the two variables (Table 5). However, in the primary dentition, the caries experience, assessed using the decayed (d), missing (m), and filled (f) index for primary teeth, for children with a combined cleft of the lip and palate (mean dmft = 1.73) was double that of children with a cleft of the lip only (mean dmft = 0.86).

All the 4-year-olds had evidence of erosion of enamel in the primary teeth (incisors and first molars) and 56% of the 12-year-olds had erosion of permanent teeth (incisors and first permanent molars).

TABLE 3 Number of Children, All Ages Combined, With Enamel Hypoplasia on Posterior or Anterior Permanent Teeth by Cleft Type*

Cleft Type	*Enamel Hypoplasia (Anterior Teeth)*	*Enamel Hypoplasia (Posterior Teeth)*
Lip	6 (8)	0 (0)
Palate	6 (7)	4 (5)
Lip and palate	4 (11)	1 (3)

* Some children may have enamel hypoplasia affecting both anterior and posterior teeth.

TABLE 4 Mean Dental Caries Prevalence by Systemic Fluoride Exposure

	Fluoride Status		
	*Full**	*Part†*	*None‡*
Number of children	N = 50	N = 16	N = 25
dmft§ (mean)	1.04	1.00	1.88
DMFT‖ (mean)	0.84	1.00	0.96
Age, y (mean)	9.1	9.0	9.1

* Full-time exposure to either fluoridated water or fluoride supplements.
† Discontinuous residence in a fluoridated area or intermittent use of fluoride supplements.
‡ Never lived in a fluoridated area nor received fluoride supplements.
§ dmft = decayed, missing, and filled index for primary teeth.
‖ DMFT = Decayed, Missing and Filled index for permanent teeth.

DISCUSSION

This study has demonstrated a relationship between the severity of the cleft (i.e., those children with clefts of both the lip and palate) and the number of teeth with hypoplastic defects. Although this is a recognized sequela of clefting, the etiology for this is unclear (Olin, 1964; Dahllof et al., 1989; Vichi and Franchi, 1995).

Research on fetuses, as well as dental casts from a postnatal sample, has concluded that the factors that led to clefting are also responsible for the enamel defects observed (Jordan et al., 1966). Microradiographic analysis of teeth adjacent to the cleft site appear to have a different mineral structure antenatally, indicating effects attributable to the clefting process or whatever was responsible for the defect (Lagarde et al., 1987). The evidence that both dentitions are involved as well as the location of the enamel defects led Malanczuk et al (1999) also to conclude that the etiology lay in the pathological process resulting in clefting. However, the presence of enamel defects in both dentitions has led other workers to implicate factors such as surgical intervention as well as the inevitable feeding difficulties leading to nutritional and metabolic upset in the process (Dixon, 1968; Ranta 1986). As in our study, Dixon (1968) also demonstrated a relationship between the site of the cleft and the prevalence of enamel defects.

The slightly higher prevalence of dental caries demonstrated in these children, by comparison with earlier studies on cleft child populations, may be due in part to the wide age range of the children. In other studies, the data are presented in discrete age bands whereas our data cover all ages from 4 to 12 years. It is difficult, therefore, to compare across the various

TABLE 5 Mean Dental Caries Experienced by Cleft Type; All Ages and Genders Combined

	Cleft Type		
Variable	*Lip*	*Palate*	*Lip and Palate*
Number of children	N = 24	N = 41	N = 26
dmft (mean)	0.86	1.20	1.73
DMFT (mean)	0.71	0.98	0.96
Age, y (mean)	9.7	8.8	8.9

studies, but the results are broadly similar. Paul and Brandt (1998) and Bokhout et al. (1997) reported data that showed a slightly lower caries experience whereas the study population of Gregg et al. (1999) had an increased caries experience as compared with our data. This may be due in part to regional differences, particularly in cases in which these reflect differing fluoridation status. Dahllof et al. (1989) were not able to attribute the significantly increased prevalence of dental caries in their study population, relative to unaffected controls, to dietary differences between study and control children.

No other studies on the oral health of children with clefts have reported on the prevalence of dental erosion, a clinical condition that has been found to be significant in other child populations (O'Brien, 1994). This study has shown that the prevalence of dental erosion, in both dentitions, is high and is another aspect that should be addressed by pediatric dentists as part of a preventive and, if necessary, treatment program for these children.

Prevention of dental disease in these patients is obviously an important issue. Dental caries and one of its sequelae—extraction of teeth—may make the orthodontic management of these children more complex. It may also hinder good speech development. A valuable tool in the pediatric dentist's armamentarium is the use of systemic and topical fluorides. The relationship between caries and systemic fluoride was not found to be as conclusive as in the Irish study (Gregg et al., 1999). However, there did appear to be benefits conferred on those that had had fluoride supplements because this study reported an inverse relationship between this form of fluoride ingestion and caries experience.

The Newcastle study is ongoing, and because of the relatively small sample size within some of the subgroups, it is too early to determine which of these differences are clinically significant.

Given the high caries experience of some children, we would advocate a more rigorous approach to the prevention of dental disease. Data from a study of similar patients in Northern Ireland would indicate that aggressive prevention by pediatric dentists makes a positive, significant difference to the oral health of these already compromised children. Not only has such contact allowed reinforcement of the preventive message but it has also allowed the identification and targeting of high-risk children for restorative care.

Early identification of enamel defects and other related dental abnormalities is important for appropriate interceptive management to ensure an optimal outcome (Vichi and Franchi, 1995).

It would appear that the pediatric dentist, in conjunction with a dietitian, should be giving specific dietary advice rather than relying predominantly on a self-help group (Fitzgerald et al., 1990). In addition, it is the role of the consultant in pediatric dentistry to ensure that these patients receive appropriate primary care—regular checkups, radiographs, oral hygiene advice, diet advice, and appropriate fluoride supplementation—and, when required, appropriate referral for secondary care.

Acknowledgments. We would like to thank the patients and their parents without whom this study would not have been possible. We are grateful for the support from staff at the Dental School and Hospital, Newcastle upon Tyne, United Kingdom, in particular the staff of Child Dental Health.

References

Bokhout B, Hofman FXWM, van Limbeek J, Kramer GJC, Prahl-Anderson B. Incidence of dental caries in the primary dentition in children with a cleft lip and/or palate. *Caries Res.* 1997;31:8–12.

Clarkson JJ, O'Mullane DM. A modified DDE index for use in epidemiological studies of enamel defects. *J Dent Res.* 1989;68:445–450.

Dahllof G, Ussisoo-Joandi R, Ideberg M, Modeer T. Caries, gingivitis and dental abnormalities in pre-school children with cleft lip and/or palate. *Cleft Palate J.* 1989;26:233–238.

Dixon DA. Defects of structure and formation of teeth in persons with cleft palate and the effect of reparative surgery on the dental tissues. *Oral Surg.* 1968;25:435–446.

Fitzgerald J, King NM, Reid CA. Parental counselling for CLP infants in Hong Kong and Newcastle upon Tyne. Presented at the Second Asian Pacific Cleft Lip and Palate Conference; 1990; Singapore.

Gregg TA, Johnson D, Pattinson KE. Efficacy of specialist care for caries in the cleft child. *Int J Paed Dent.* 1999;9(suppl):61.

Jordan R, Bertram SK, Marshall Neptune C. Dental abnormalities associated with cleft lip and/or palate. *Cleft Palate J.* 1966;3:22–55.

Lagarde A, Kerebel B, Ajacques JC. Structure, ultrastructure and microanalysis of the enamel of teeth near maxillary clefts. *Ann Pathol.* 1987;7:113–121.

Lagarde A, Kerebel B, La Cabellec MT. Anomalies of enamel formation in subjects with maxillary clefts. *Bull Group Int Rech Sci Stomatol Odontol.* 1989;32:191–197.

Malanczuk T, Opitz C, Retzlaff R. Structural changes of dental enamel in both dentitions of cleft lip and palate patients. *J Orofac Orthop.* 1999;60:259–268.

O'Brien M. Children's Dental Health in the United Kingdom 1993. London, England: Her Majesty's Stationary Office; 1994.

Olin WH. Dental anomalies in cleft lip and cleft palate patients. *Angle Orthod.* 1964;34:119–124.

Paul T, Brandt RS. Oral and dental status of children with cleft lip and/or palate. *Cleft Palate J.* 1998;35:329–332.

Ranta R. A review of tooth formation in children with cleft lip/palate. *Am J Orthod Dentofacial Orthop.* 1986;90:11–18.

Rivkin CJ, Boulton SE, Hathorn IS, Crawford PJ. Measurements of caries experience and dental attendance in cleft patients. *Int J Paediatr Dent.* 1999; 9(suppl):62.

Vichi M, Franchi L. Abnormalities of the maxillary incisors in children with cleft lip and palate. *ASDC J Dent Child.* 1995;62:412–417.

Wong FW, King NM. The oral health of children with clefts—a review. *Cleft Palate J.* 1998;35:248–54.