PEDIATRIC DENTISTRY/Copyright © 1988 by
The American Academy of Pediatric Dentistry
Volume 10, Number 2

Dental development in hemifacial microsomia I. Eruption and agenesis

Maria Farias, DDS Karin Vargervik, DDS

Abstract

The extent of anomalies and developmental aberrations in hemifacial microsomia (HFM) have not yet been fully delineated. Thus, the developmental pattern of the dentition in patients with HFM remains unclear. This study was conducted to investigate the development of the dentition in subjects with varying degrees of HFM and to compare the affected side with the unnaffected side and with data from control samples. Detailed tracings of the complete dental profile of 60 patients with HFM between the ages of 6 and 24 years were obtained from panoramic radiographs. The degree of development of each tooth was compared to Nolla's stages of tooth calcification, and developmental scores were assigned. The present study demonstrates that in individuals with HFM: (1) tooth development is frequently delayed on the affected side; (2) the mandibular third molar is more frequently missing on the affected than on the unaffected side; and 3) the incidence of missing teeth is higher than in unaffected subjects. A possible linkage between tooth development aberrations and the pathogenesis of HFM is discussed.

Hemifacial microsomia (HFM) is a predominantly unilateral malformation of craniofacial structures that develop from the first and second branchial arches. Characteristic features of HFM are the underdevelopment of the mandibular condyle and ramus, the zygomatic arch and malar bone, and associated soft tissues.[1] Asymmetric tooth maturation and tooth agenesis also have been found in patients with HFM (Rushton 1953; Loevy and Shore 1985). However, the extent of dental anomalies in patients with HFM has not yet been fully investigated, and the developmental pattern of the dentition in these patients remains unclear.

This study was conducted to determine whether a difference exists in the development of the dentition on the affected side compared with the unaffected side in patients with varying degrees of HFM. In addition, the development of the dentition in patients with HFM was compared to that in the general population.

[1] Poswillo 1973; Vargervik and Miller 1983; Vargervik et al. 1986; Zbynek 1986.

Materials and Methods

Tooth development and agenesis were studied in 60 patients (24 boys, 36 girls) with HFM treated at the Center for Craniofacial Anomalies, University of California, San Francisco. They ranged in age from 6 to 24 years and were divided into four groups (Fig 1): younger than 10 (N = 12); 10-13 (N = 35); 14-17 (N = 7); and older than 17 (N = 6). Patients younger than 14 years were excluded from the assessment of third molar agenesis because 14 years is considered a cut-off age for determining if this tooth will develop (Banks and Harry 1934; Garn and Lewis 1962). Patients were grouped according to the extent of mandibular deformity (Chierici 1983): type I — absence of condylar cartilage and disc; type II — no condylar head or neck on condylar process; type III — ankylosis or syndesmosis of joint; type IV — presence of coronoid process only; and type V — coronoid and condylar processes missing. The patient sample was composed as follows: type I — 26 (43%); type II — 3 (5%); type III — 4 (7%); type IV— 17 (28%); and type V — 10 (17%).

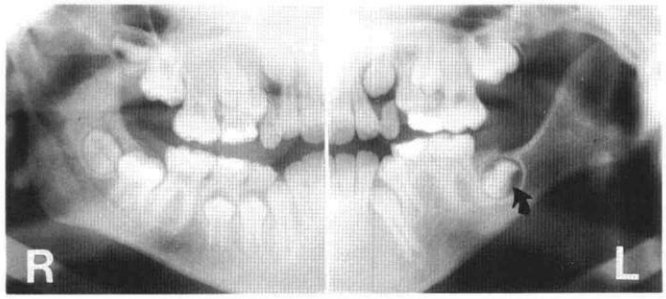

FIG 1. Comparison between delay in tooth development on the affected (A) and the unaffected side (N/A) in subjects with HFM grouped by age ($P < 0.001$, significantly different).

Detailed tracings of the complete dental profile of each patient were obtained from panoramic radiographs (Fig 2 — next page). The degree of development of each tooth was evaluated and compared with Nolla's stages of tooth calcification (Nolla 1960), and a developmental score was assigned. Data were com-

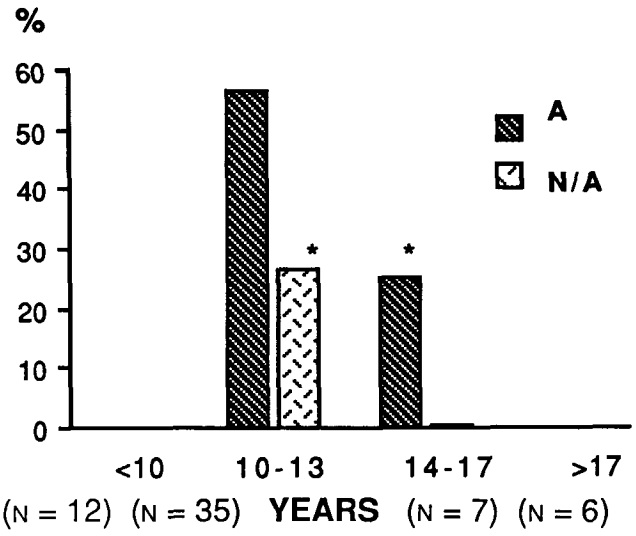

FIG 2. Panoramic radiograph of a 9-year-old girl illustrates agenesis of mandibular left third molar and mandibular left second premolar on the affected side and delayed tooth development of the permanent mandibular left second molar on the affected side *(arrow)*.

pared by chi-square ($\chi 2$) with Yate's correction for continuity.

Results

Delayed development was found in 41 permanent teeth of the affected side in our subjects with HFM. The mandibular second and third molars were the most frequently affected teeth (Table 1). Thirty of the 60 subjects had delayed tooth development on the affected side. Delayed tooth development was more often found on the affected side of subjects with types III, IV, or V

TABLE 1. Delayed Development of Permanent Mandibular Teeth on the Affected Side in Subjects with HFM

	(N)	Per cent
First premolar	2	4.88
Second premolar	4	9.76
First molar	2	4.88
Second molar	18	43.90
Third molar	15	36.58
Total	41	100

than in subjects with type I or II malformation. Sixty-five per cent of subjects with type I malformation showed equal tooth development on both sides and 35% showed less tooth development on the affected side. All patients with type II malformation had equal tooth development on both sides. In contrast, 100% of the patients with type III, 71% with type IV, and 50% with type V malformation had delayed tooth development on the affected side (Table 2).

Delayed tooth development was not found in subjects younger than 10 years. However, 17 of the 30 subjects between 10 and 13 years (56.6%) showed delayed tooth development on the affected side. The difference in tooth development between affected and unaffected side was statistically significant ($P < 0.001$). The incidence of delayed tooth development decreased after age 13, and teeth on the affected and on the unaffected side reached a similar degree of development after age 17 (Fig 1).

TABLE 2. Tooth Development on the Affected (A) and the Non-Affected Sides in Subjects with HFM

		Equal Development		Less Development (A)	
Type	(N)	(N)	Per cent	(N)	Per cent
I	26	17	65	9	35
II	3	3	100	0	0
III	4	0	0	4	100
IV	17	5	29	12	71
V	10	5	50	5	50
Total	60	30		30	

Unilateral absence of teeth was more often found on the affected (28 teeth) than on the unaffected side (3 teeth) ($P < 0.001$). Eighteen teeth were absent bilaterally. The most frequently missing teeth were the mandibular third molar and the mandibular second premolar (Table 3). Missing teeth other than third molars were found in 25% of the subjects with HFM. The percentage of missing teeth other than third molars was higher on the affected than on the unaffected side ($P < 0.001$). A missing third molar was more often found on the affected than on the unaffected side (40.1 vs. 13.3%, $P < 0.001$, Fig 3 — next page). The incidence of missing teeth was higher in females than in males.

TABLE 3. Frequency of Missing Teeth in Subjects with HFM

Permanent Dentition	Unilateral A	Unilateral N/A	Bilateral
Maxillary teeth			
Lateral incisor	0	0	1
First premolar	1	0	1
Second premolar	1	1	1
First molar	1	0	0
Second molar	1	0	0
Third molar	1	0	0
Mandibular teeth			
Lateral incisor	2	0	0
First premolar	1	0	0
Second premolar	4	1	4
Second molar	2	0	0
Third molar	14	1	11
Total	28	3*	18

* $P < 0.001$ significantly different from A.

Discussion

This study shows that tooth development is significantly delayed on the affected side in patients with HFM. Fifty per cent of our subjects with HFM had delayed tooth development on the affected side. This finding is different from the observation of Loevy and

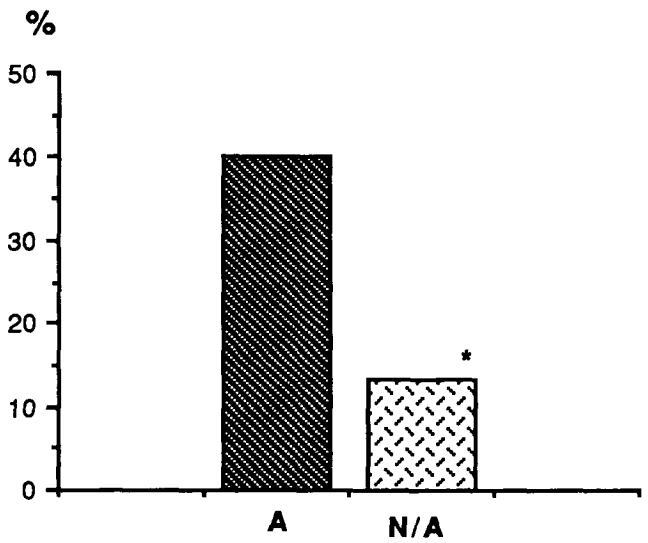

FIG 3. Missing third molars on the affected (A) and the unaffected side (N/A) in subjects with HFM ($P < 0.001$, significantly different).

Shore (1985) who reported that only 20 of their 81 patients with HFM (24.7%) had delayed tooth development on the affected side. The difference between Loevy and Shore's and our results might be due to the fact that half of our patients had more severe types of HFM (types III, IV, and V malformation); whereas, 64% of Loevy and Shore's patients were classified as class I (Pruzansky 1969). Thus, it appears that the incidence of delayed tooth development in subjects with HFM is proportional to the extent of mandibular deformity. This assumption is supported by our finding that delayed tooth development and agenesis are more frequent in subjects with HFM type III, IV, or V than in patients with type I or II anomalies. It is likely, therefore, that the pathologic processes that result in HFM also cause disturbances in the development of the dental lamina, and that the severity of the embryonic insult determines the degree of mandibular lesion and the incidence of both delayed tooth development and missing teeth.

Our data show that the highest incidence of delayed tooth development in subjects with HFM occurs between 10 and 13 years of age. This fact may reflect a delay of the active eruptive phase that accompanies the adolescent growth spurt (Darling and Levers 1975; Steedle and Proffit 1985).

The delayed tooth development on the affected side in subjects with HFM contrasts with the approximately equal rate of growth of permanent dentition on both sides of the mouth in the normal population (Banks and Harry 1934). Likewise, the overall incidence of missing teeth other than third molars in subjects with HFM is higher than that found in unaffected individuals. In our sample, 25% of the patients had missing teeth other than third molars, whereas the incidence of missing teeth other than third molars in unaffected subjects has been reported to be 4-6.5% (Horowitz 1966; Silverman and Ackerman 1979). Finally, the incidence of a missing third molar on the affected side in subjects with HFM is more than twice that reported in unaffected individuals (40.1 vs. 16%), but the percentage of a missing third molar on the unaffected side is not significantly different from that found in the normal population (13.3 vs. 16%, Garn 1963). These differences in dental development between subjects with HFM and unaffected individuals provide additional evidence to support the hypothesis that the pathogenesis of HFM also alters dental development on the affected side.

The etiology of HFM is not yet clearly understood, and the cause of the high incidence of missing teeth and delayed tooth development on the affected side in patients with HFM is unknown. It has been implied that a vascular lesion is a major factor in the etiology of first and second branchial arch deformities (Poswillo 1973). Hemorrhage of the stapedial artery in the mouse and in the monkey at the time of odontogenic epithelium formation produces similar otomandibular malformations to those seen in patients with HFM (Poswillo 1973; ten Cate 1980). Therefore, deficient vascularization at an early stage of cell differentiation and migration might lead not only to bone, cartilage, and muscle underdevelopment, but also to delay in the development of the dental lamina.

Clinical observations indicate that other aspects of dental development such as tooth size and morphology might also be abnormal in patients with HFM. These parameters of dental development will be subjected to further studies.

Conclusions

In subjects with HFM: (1) tooth development is frequently delayed on the affected side; (2) delayed tooth development is more frequent during the second phase of active eruption; (3) the mandibular third molar is the most frequently missing tooth and is more often missing on the affected than on the unaffected side; and (4) the incidence of missing teeth is higher than that reported in unaffected individuals. These aberrations in the dental development might be related to the same pathogenic processes that cause HFM.

This study was supported by NIH Grant DE04940.

Dr. Farias is an assistant professor, Central University of Venezuela, and Dr. Vargervik is a professor of growth and development and director, Center for Craniofacial Anomalies, University of California at San Francisco. Reprint requests should be sent to: Dr. Karin Vargervik, S-747 Center for Craniofacial Anomalies, University of California, San Francisco, CA 94143-0442.

Banks HV, Harry B: Incidence of third molar development. Angle Orthod 4:223-33, 1934.

Chierici G: Radiologic assessment of facial asymmetry, in Treatment of Hemifacial Microsomia, Harvold EP, Vargervik K, Chierici G, eds. New York; Alan R. Liss Inc, 1983 pp 57-87

Darling AI, Levers BG: The pattern of eruption of some human teeth. Arch Oral Biol 20:89-96, 1975.

Garn SM, Lewis AB: The relationship between third molar agenesis and reduction in tooth number. Angle Orthod 32:14-18, 1962.

Garn SM: Third molar polymorphism and its significance to dental genetics. J Dent Res 42:1344-63, 1963.

Horowitz JM: Aplasia and malocclusion: a survey and appraisal. Am J Orthod 52:440-52, 1966.

Loevy HT, Shore SW: Dental maturation in hemifacial microsomia. J Craniofac Genet Dev Biol 1:267-72, 1985.

Nolla CM: The development of the permanent teeth. J Dent Child 27:254-66, 1960.

Poswillo D: The pathogenesis of the first and second branchial arch syndrome. Oral Surg 35:302-28, 1973.

Pruzansky S: Not all dwarfed mandibles are alike. Birth Defects 5:120-29, 1969.

Rushton MA: Asymmetry of tooth size in congenital hypoplasia of one side of the body. Br Dent J 95:309-11, 1953.

Silverman NE, Ackerman JL: Oligodontia: a study of its prevalence and variation in 4032 children. J Dent Child 46:470-77, 1979.

Steedle JR, Proffit WR: The pattern and control of eruptive tooth movements. Am J Orthod 87:56-66, 1985.

ten Cate AR: Embryology of the head, face, and oral cavity, in Oral Histology, ten Cate AR, ed. St Louis; CV Mosby Co, 1980 pp 18-45.

Vargervik K, Ousterhout D, Farias M: Factors affecting long-term results in hemifacial microsomia. Cleft Palate J 23:53-68, 1986.

Vargervik K, Miller A: Assessment of facial and masticatory muscles in hemifacial microsomia, in Treatment of Hemifacial Microsomia, Harvold EP, Vargervik K, Chierici G, eds. New York; Alan R Liss Inc, 1983 pp 113-32.

Zbynek S: Craniofacial changes in hemifacial microsomia. J Craniofac Genet Dev Biol 6:151-70, 1986.

Occupational exposure to AIDS

Health care workers (HCWs) are at minimal risk for infection from the human immunodeficiency virus (HIV) from occupational exposure to patients with AIDS or AIDS-related conditions, a recent San Francisco study says.

Most of the HCWs had been exposed to patients with AIDS for at least one year before enrolling in the study. When tested at the time of enrollment, none of the 270 subjects chosen had antibodies for HIV or were in a high-risk group for the disease.

In their conclusions, the researchers noted that the subjects in the study "represent one of the most highly exposed cohorts of HCWs in the world." The absence of HIV transmission to these individuals "is extremely reassuring and adds significant power" to previous studies of health care workers.

The researchers added that "the relatively poor compliance with recommended infection control practices reported by the study subjects is disturbing" despite the absence of HIV transmission. They recommended additional efforts to encourage compliance with infection control guidelines.