



Multiple congenitally missing primary teeth: report of a case

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Abstract

The prevalence, possible etiological factors and management of multiple missing primary teeth was briefly reviewed. Oligodontia of the primary dentition is a rare finding. This paper reports a rare case of multiple missing (n=9) primary teeth in a 3-year-old male of Asian origin. (*Pediatr Dent* 24:149-152, 2002)

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The literature is replete with articles describing the congenitally missing permanent teeth; however little has been written describing the number, frequency and location of congenitally missing primary teeth.¹⁻¹³ Agenesis is rare in the primary dentition.¹⁴ Indeed, the reduction in number of teeth is concomitant with the reduction in the size of the jaw in human evolution and believed to be a continuing evolutionary trend.

Nevertheless, causes of missing teeth, particularly anodontia – an extreme expression of oligodontia – are very rare and challenging to the clinician.¹⁵ Scientific analysis of the congenital absence of teeth in humans dates back to early 1900s.¹⁶ Clayton, in 1956,¹⁷ collected data from 3,557 full-mouth intraoral radiographs. He reported that 214 children in this sample, ranging in age from three to 12 years, had a total of 433 congenitally missing teeth. Six percent of the 214 children were missing more than one tooth. The incidence of hypodontia in the primary dentition varies ranging from 0.5% among the Swedish children to 1.0% among the Caucasians.¹⁸ However, a higher incidence of 5.0% in the primary dentition of Japanese children has been reported.¹⁹

Brabant, in 1967,²⁰ stated that “hypodontia of the deciduous dentition” has always been infrequent. He reported a frequency not exceeding two to six per 1,000 children. Brook, in 1974,²¹ investigated 741 British children aged three to five years, and 1,115 children aged 11 to 14 years. He found the prevalence of hypodontia to be less than 1% with no sex predilection.

Teeth may be missing due to prior extraction or failure to develop or erupt.^{18,22} The most common dental anomaly occurring in association with congenital absence of the

permanent lateral incisor/second premolar is the absence of other teeth.²³

Hypodontia among primary teeth is a relatively rare occurrence with a prevalence of less than 1%. Hypodontia usually affects maxillary lateral incisors and mandibular central and lateral incisors.²⁴

Witkop, in 1965,²⁵ reported that in one Swiss isolate, 33% of the population was found to be missing maxillary lateral incisors. Genealogic studies proved that all the affected individuals were descendants of one man who had shown the characteristic in the 18th Century. The classical family study of oligodontia was completed by Grahen in 1956²⁶ on parents and siblings of 171 persons with hypodontia. He concluded by stating that in the majority of cases, oligodontia is mainly determined by a dominant autosomal gene pattern with incomplete penetrance of the trait and variable expressivity.

Case report

The patient, a three-year-old Asian male, was referred to the department of Pedodontics and Preventive Dentistry, College of Dental Sciences, with a chief complaint of missing primary teeth. The mother gave a history of consanguineous marriage. The family history regarding the absence of teeth was not significant. Even the siblings had no such history.

An intraoral examination revealed the presence of maxillary right and left primary central incisor, maxillary right and left primary first molar, mandibular right central incisor and mandibular right and left primary first molars. There was also the presence of ankyloglossia (tongue-tie) and high labial frenal attachments in the maxillary and mandibular



Fig 1.

arches. The alveolus present was also very thin. The teeth present were of normal size, shape and color (Fig 1).

Intraoral periapical radiographs revealed the presence of maxillary central incisor tooth buds. There was congenital absence of the maxillary right and left primary lateral incisors, maxillary right and left primary canines (cuspids), mandibular left primary central incisor, mandibular right and left primary lateral incisors and canines (cuspids). The radiographs also revealed unerupted maxillary and mandibular second primary molars (Fig 2).

Discussion

This case is interesting for several reasons. First, there are very few cases reported in the literature regarding congenital absence of primary teeth, and, in this case, there are a total of nine missing primary teeth, which in itself is very rare. Secondly radiographic evidence of this case showed the presence of the maxillary permanent central incisors tooth buds.

At the age of three years, usually the permanent maxillary central and lateral incisor and mandibular central and lateral incisors tooth buds should be present.²⁷ But in this case, only the permanent maxillary central incisor tooth buds are present and the other tooth buds are absent. They could be formed later or they can be congenitally missing. Thirdly, the patient exhibited ankyloglossia and high frenal attachments with very thin alveolar ridges. Certainly this patient has serious deficiencies and will require long term preventive and prosthodontic management.

The most common developmental problem with this type of clinical picture is ectodermal dysplasia. Ectodermal dysplasia can be classified mainly as:²⁸ (1) hair defect, (2) tooth defect, (3) nail defect, (4) sweating defect.

This can be classified again into many types based on the characteristic features, and few conditions associated with the absence of teeth are:

1. *Hypohidrotic ectodermal dysplasia/Anhidrotic ectodermal dysplasia*: Is characterized by partial/complete absence of sweat glands, hypotrichosis and hypodontia. In this condition, primary/permanent teeth may be entirely absent/few may be present. Incisors and canines are

conical and pointed.²⁹ The conical, pointed teeth are the key feature of the syndrome and may be the only obvious abnormality.³⁰

2. *Tricho-onychodental dysplasia*: A rare syndrome characterized by taurodontic molars, defective enamel and dentin dysplasia. There are few teeth, and widely spaced and deciduous teeth tend to persist. Nails are thin with longitudinal striations and cracks.³¹
3. *Fried's tooth and nail syndrome*: In this condition, hair is fine and short, the teeth are few and peg shaped and the nails are thin and dystrophic.³²
4. *Hypodontia and nail dysgenesis*: Is characterized by the presence of few teeth which are conical and widely shaped. Nails are small, dystrophic/spoon shaped.³³⁻³⁶
5. *Odontomicronychial ectodermal dysplasia*: This condition is characterized by precocious eruption and shedding of deciduous dentition, precocious eruption of secondary dentition with short, rhomboid roots and short, thin, slow-growing nails.³⁷
6. *Odonto-trichomeelic syndrome*: Characterized by severe hypotrichosis, few, small, conical teeth and hypoplastic or absent aerolae. There is cleft lip and extensive tetramelic dysplasia. Growth is retarded.³⁸⁻⁴¹

However, this patient does not have peg-shaped teeth, dry, scaly skin, straw-like hair, frontal bossing, wide spaced eyes or any problem with body-heat regulation. He appears to be a perfectly normal young child, with no known family history of missing teeth. The features in this patient also are not consistent with any specific forms of ectodermal dysplasia.

Conclusion

This is a classical case of oligodontia with multiple missing primary teeth. It is a rare case which exemplifies the need for a thorough diagnosis, when primary teeth are missing. These conditions being rare may be the first evidence of a much more severe problem in the permanent dentition. Early recognition permits long-range planning, implementation of aggressive preventive activities and routine developmental observations.

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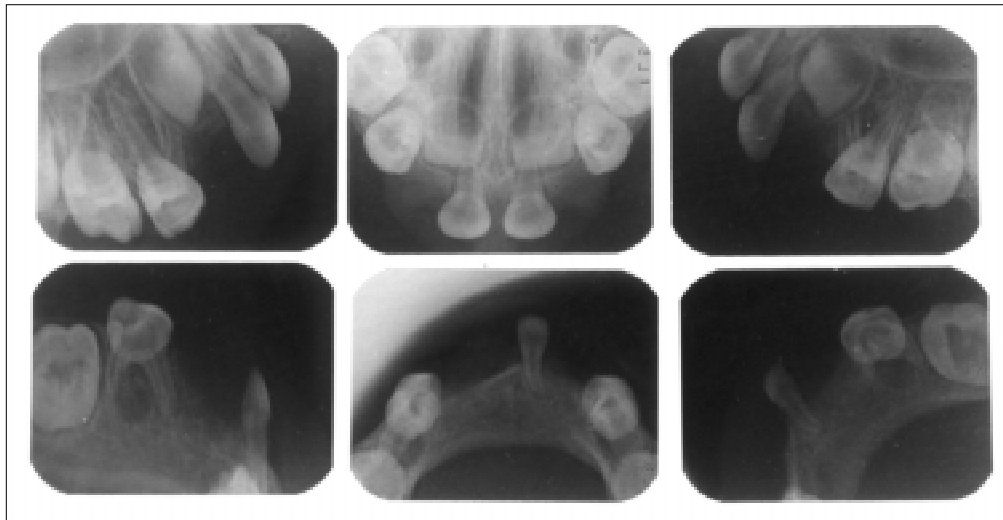


Fig 2.

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ABSTRACT OF THE SCIENTIFIC LITERATURE



ASTHMA MORBIDITY AFTER THE SHORT-TERM USE OF IBUPROFEN IN CHILDREN

Aspirin and other nonsteroidal anti-inflammatory drugs (NSAIDs) can trigger acute bronchospasm in sensitive adults and children. Because of its known association with Reye syndrome, aspirin is rarely given to children today. As a result, potential sensitivity to aspirin or other NSAIDs may not be recognized in young children with asthma. The authors tested the hypothesis that among children without a history of aspirin sensitivity, the use of ibuprofen suspension for fever control increases the risk of acute bronchospasm and other morbidity from asthma.

A randomized, double-blind, clinical trial was conducted. A total of 1,879 patients who had asthma and a febrile illness were randomly assigned to receive suspensions of either acetaminophen (12 mg/kg, 632 subjects) or ibuprofen in 1 of 2 dosages (5 mg/kg, 636 subjects or 10 mg/kg, 611 subjects) and were followed for 4 weeks. Rates of hospitalization and outpatient visits for asthma during follow-up were compared. Rates of hospitalization for asthma did not vary significantly by antipyretic assignment. However, the risk of an outpatient visit for asthma was significantly lower in the ibuprofen group, which has the relative risk of 0.56 (95% confidence interval: 0.34-0.95). The risk of an outpatient visit for asthma did not vary by assigned ibuprofen dose (i.e., 5 mg/kg, 10 mg/kg).

The results indicated that compared with acetaminophen, ibuprofen may reduce risks of asthma morbidity. It remains to be determined whether the observed difference in morbidity is attributable to increased risk after acetaminophen or a decrease after ibuprofen use. These data provide evidence of the relative safety of ibuprofen use in children with asthma.

Comments: Ibuprofen has been shown to preserve pulmonary function better than placebo in children with cystic fibrosis and mild lung disease. This report provides a scientific support of using ibuprofen in children with asthma.

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Lesko, SM, Louik C, Vezina RM, Mitchell AA. Asthma morbidity after the short-term use of ibuprofen in children. Slone Epidemiology Unit, School of Public Health, Boston University School of Medicine, Boston, Massachusetts *Pediatrics* 109(2): e20, February 2002.

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