## Congenitally missing permanent lateral incisors in conjunction with a supernumerary tooth: case report

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upernumerary teeth, or hyperdontia, is defined as an excess number of teeth when compared with the normal dentition. Classification is either based on time of appearance (predeciduous, similar to permanent, postpermanent, and complementary) or according to positions in the dental arch (mesiodens, paramolars, postmolars, or impacted).1

Various studies reporting the prevalence of supernumerary teeth and congenitally missing teeth have produced a range of occurrence rates.<sup>2-8</sup> Such variation may be attributed to study design and racial differences among the groups examined. Niswander reported a 3.4% frequency of supernumerary teeth among 4150 Japanese subjects.<sup>2</sup> In a study of 12-year-old Hong Kong Chinese, Davis found a 2.7% prevalence of supernumerary teeth.3 From panoramic radiographs of 5- to 13-yearold Finnish children, Haavikko reported a 1.6% occurrence of supernumerary teeth.4 This was similar to a report by Ravn and Nielsen who found a 1.3% frequency among 7- to 10-year-old schoolchildren from Copenhagen.<sup>5</sup> These reports seem to suggest a racial variation in the prevalence of supernumeraries, with a higher frequency in the Asian population.

The occurrence of supernumerary teeth in the primary dentition is a less common finding, one-fifth of that seen in the permanent dentition.9 The subsequent occurrence of supernumerary teeth in the permanent dentition of children with primary dentition supernumeraries is estimated at approximately 30%.10,11 Studies demonstrate a male predilection ranging from 2:1 in most populations 12, 13 to a 6.5:1 ratio in Hong Kong Chinese.4

Congenital absence of teeth (hypodontia) is the most common developmental dental anomaly in man. 14 Congenitally absent teeth are more common in the permanent dentition and have a prevalence ranging from 3.7 to 9% (excluding the third molars) with a female to male ratio of 3:2.15 Hypodontia in the primary dentition is rare, generally appearing in the incisor region and often associated with missing succedaneous teeth. 16 Individuals with congenitally absent teeth also frequently have microdont, conical or tapered teeth, reduced alveolar development, an increased freeway space, and retained primary teeth. Hypodontia is commonly accompanied by malformation of the corresponding tooth on the contralateral side. 17, 18

The terms, "concomitant hypodontia and hyperodontia"19 and "oligo-pleiodontia"20 have been used to describe the condition in which developmental absence of teeth and supernumerary teeth are present in the same individual. Only a few case reports of this rare condition exist in the literature. Two describe congenitally missing maxillary lateral incisors and presence of a midline supernumerary 19, 21 Three others report missing maxillary lateral incisors associated with maxillary supernumerary premolars<sup>20, 22, 23</sup> The purpose this case report is to describe the coexistence of congenitally missing maxillary lateral incisors in the presence of a supernumerary tooth in the mandibular first molar area.

## Case report

A healthy Hispanic male, aged 14 years 11 months, was referred to the department of pediatric dentistry, with a chief complaint of maxillary anterior dental spacing. His medical history was unremarkable and dental history revealed root canal therapy to the right maxillary permanent central incisor and regular routine dental care. A family history of hereditary tendencies to supernumerary or congenitally missing teeth was negative.

The intraoral examination revealed healthy soft tissues in the presence of good oral hygiene practices. He had a partial permanent dentition, a 60% overbite, and Class I molar relationship. Maxillary anterior spacing was present with a 2-mm midline diastema and a retained right primary lateral incisor (Fig 1). Both permanent maxillary lateral incisors were clinically absent



Fig 1. Intraoral photograph showing maxillary anterior dental spacing with maxillary right primary lateral incisor retained.

central incisors with a 0.15–1.90% prevalence in the Caucasian population. <sup>25,26</sup> Supernumerary teeth also may be found in the palate, the incisive suture, and in other areas, such as the nasal cavity, the ophthalmic conchae, and the maxillary sinus. To date, there are two cases where a supernumerary tooth has been found situated between the orbit and the brain. <sup>27,28</sup>

Causes of supernumerary teeth include phylogenetic reversion (atavism) to extinct primates, <sup>29</sup> split in the tooth bud (dichotomy theory), <sup>30</sup> and locally conditioned hyperactivity of the dental lamina. <sup>31</sup> In addition, genetic factors have been associated with supernumeraries and include cleidocranial dysostosis, cleft lip and cleft palate, and Gardner's syndrome. <sup>12, 26, 31, 32</sup>

Fig 2. Panoramic film showing retained right maxillary primary lateral incisor, without evidence of right or left maxillary permanent lateral incisors. Coronal development of a supernumerary tooth in the apical region of the right mandibular first molar is visible.

with no history of traumatic avulsion or extraction. No carious lesions were evident and previously placed sealants and restorations presented in good condition. The maxillary right central incisor had a minor fracture and was discolored.

A panoramic radiograph revealed bilateral absence of the permanent maxillary lateral incisors (Fig 2). This was confirmed on a subsequent maxillary anterior occlusal film. The panoramic radiograph also demonstrated the presence of a molariform supernumerary tooth developing somewhat apical to the mandibular right first permanent molar. The crown appeared well developed with incomplete root formation. Third molar development was absent in both jaws.

## Discussion

Supernumerary teeth may be found in various locations in the jaws. Approximately 90–98% of all supernumeraries occur in the maxilla with a predilection for the premaxilla. <sup>12, 13, 24</sup> The most common type of supernumerary tooth is the mesiodens, located between the

According to previous reports30, 33 80-93% of the supernumeraries are associated with pathologic changes. Foremost among such sequelae are delayed eruption and/or displacement of the adjacent permanent teeth. Occasionally supernumeraries may lead to the development of dentigerous or primordial cysts, cause root resorption or rotation of the adjacent teeth, or erupt into the nasal cavity.31

After third molars, mandibular second premolars are the teeth most commonly missing, followed by either the maxillary lat-

eral incisor or the maxillary second premolar.<sup>3, 4, 17</sup> The Chinese population differs in that the mandibular incisors are the most frequently missing tooth, followed by maxillary second premolars and maxillary lateral incisors.<sup>5</sup> When a third molar is congenitally missing, the occurrence of hypodontia elsewhere in the permanent dentition was found to be up to 13 times greater than normal.<sup>34</sup> In the case reported here, it is interesting to note that all third molars buds were absent.

Congenital absence of teeth may arise from several causes including physical obstruction or disruption of the dental lamina, space limitation, functional abnormalities of the dental epithelium, or failure of initiation of the underlying mesenchyme.<sup>35</sup> It is well known that congenital absence of teeth may result from genetic factors although the modes of inheritance remain unclear.<sup>36</sup> To date, congenitally missing teeth have been reported in association with more than 50 syndromes of the head and neck.<sup>37</sup> The most common dental anomaly occurring in association with congenital absence of the permanent lateral incisor or second pre-

molar is the absence of other teeth. Other sequelae include disturbances in spacing, tooth eruption, and exfoliation.38

The simultaneous presence of supernumerary teeth and the congenital absence of other teeth is very rare. To our knowledge, this is the first such case of congenitally missing maxillary permanent lateral incisors in association with mandibular molar hyperdontia. Mercer (1970) estimated that the probability of these anomalies coexisting is between 8 and 15 per 10,000.<sup>23</sup> Rose (1974) reported a frequency of 13 in 10,000 patients.39 Gibson (1979) found 20 cases in 4598 orthodontic patients, a prevalence of 1 in 230.40 The etiology of these anomalies coexisting is unknown. Disturbances in migration, proliferation, and differentiation of the neural crest cells and interactions between the epithelial and mesenchymal cells during the initiation of odontogenesis have been suggested.37,41

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