Congenitally missing maxillary primary canines: report of three cases

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Summary. *Background.* Hypodontia is uncommon in the primary dentition and lateral incisors are the most commonly missing primary teeth in the patients affected. Studies have found that hypodontia in the primary dentition involving only maxillary canine is rare.

Case reports. Three cases of hypodontia in the primary dentition involving only maxillary canine are presented. Two of them were affected by hypodontia and hyperdontia in the permanent dentitions respectively, yet all the permanent canines developed normally in these three cases.

Conclusion. In children with congenitally missing maxillary canines, the permanent dentitions may show diverse anomalies in tooth numbers, ranging from hypodontia to hyperdontia.

Introduction

Hypodontia is the congenital absence of one or more teeth from the dentition. Recent reports have shown that the prevalence of hypodontia in the permanent dentition (third molars excluded) is about 4.5-7.4% in Caucasians, and that the most commonly missing tooth is the mandibular second premolar [1,2]. Compared with the situation in the permanent dentition, hypodontia in the primary dentition is more rare and its prevalence has been found to be 0.5% or less in several studies [3–8].

Lateral incisors are the most commonly missing teeth in the primary dentition and studies involving 927 Icelandic children [6] and 750 Belgian children [8] showed that lateral incisors were the only missing teeth in the primary dentition. It has also been shown that maxillary lateral incisors were more commonly found missing than mandibular lateral incisors. In a study of 1141 Finnish children, seven maxillary lateral incisors and four mandibular lateral incisors were found missing [9], whereas in the study of 1300 Saudi children, there were four maxillary lateral incisors and one mandibular lateral incisor missing [10]. In all the above four studies [6,8–10], not a single case of congenitally missing primary canine was found. In a study on the pattern of agenesis in 193 children with hypodontia in the primary dentition, Daugaard-Jensen and co-workers further confirmed that congenitally missing primary molars, canines, and maxillary central incisors were rare [11]. This article reports the dental findings of three children with congenitally missing maxillary primary canines.

Case reports

Case 1

The first case was a 6-year-old Chinese boy who had no previous dental experience. His medical history was unremarkable, and there was no history of supernumerary or congenitally missing teeth in his family. Intraorally, he was in early mixed dentition with maxillary left and mandibular left and right primary central incisors exfoliated. Both mandibular permanent central incisors have erupted. The two maxillary primary canines were missing clinically, and a premaxillary supernumerary tooth has erupted (Fig. 1). His mother confirmed the maxillary primary canines were congenitally missing. Caries was found at the occlusal surface of the mandibular left primary second molar. An orthopantomograph taken to check for other dental anomalies showed all permanent teeth developing (third molars excluded) (Fig. 2). The maxillary left permanent central incisor was delayed in eruption. The carious lesion was subsequently restored and the supernumerary tooth was extracted

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Fig. 1. Frontal view of case 1 showing congenitally missing 53 and 63. A supernumerary tooth has erupted in the space of the maxillary left central incisor.

under local anaesthesia. At the 1 year recall, all maxillary permanent incisors were still unerupted. Radiographic examination showed that the left central incisor has moved incisally but was still delayed when compared with the adjacent teeth. The left central incisor remained unerupted 20 months after extraction of the supernumerary tooth, while the adjacent incisors had erupted at this time. The unerupted incisor was surgically exposed, and the tooth started to erupt steadily. The boy was then followed up regularly for his dental development.

Case 2

The second case was a 6-year-old Chinese boy with unremarkable medical history and no previous dental history. There was no family history of super-



Fig. 2. Radiograph of case 1 showing presence of all permanent teeth (except third molars) and a premaxillary supernumerary tooth.

numerary or congenitally missing teeth. Intraorally, the boy was in an early mixed dentition with maxillary right permanent first molar and both mandibular permanent central incisors erupted. The maxillary right primary canine was congenitally missing, which was confirmed by his mother. Caries was seen on the proximal surfaces of five primary molars. An orthopantomograph taken to check for other dental anomalies showed congenitally missing maxillary left and both mandibular permanent second premolars (Fig. 3). Roots of the mandibular left primary second molar have been resorbed by the erupting permanent first molar. The carious lesions were subsequently restored and the boy was reviewed regularly to monitor his dental development.

Case 3

The third case was a 6-year-old Chinese boy when first seen by the authors. His medical and family history



Fig. 3. Radiograph of case 2 showing congenitally missing 53, 25, 35, and 45.



Fig. 4. Periapical radiograph of case 3 showing congenitally missing 63.

was unremarkable. He was in primary dentition with congenitally missing maxillary left primary canine. A periapical radiograph showed the presence of the corresponding permanent canine (Fig. 4). The boy was followed up annually and an orthopantomograph taken at 9 years of age showed the presence of all permanent teeth (third molars excluded).

Discussion

The cases in this report were considered uncommon as maxillary primary canines were seldom found missing congenitally. Ravn found 25 cases of hypodontia in the primary dentition of 4564 3-yearold children: 17 of them had missing maxillary lateral incisors, seven had missing mandibular lateral incisors, and only one case was found to have a missing mandibular primary canine [4]. Whittington and Durward examined 1680 5-year-old children and found only one congenitally missing mandibular primary canine [7]. However, in both studies not a case of missing maxillary primary canines was found.

Daugaard-Jensen and co-workers studied the pattern of agenesis in 193 children with hypodontia in the primary dentition [11]. Maxillary lateral incisors were the most frequently missing teeth followed by mandibular lateral incisors. Congenitally missing primary molars, canines and maxillary central incisors were found to be rare in their study. There were only four missing maxillary primary canines out of 329 missing teeth [11]. In a follow-up study of 33 hypodontia cases in the primary dentition with complete radiographic records of both dentitions, only two cases had missing maxillary primary canines [12]. Both of them had five or more missing teeth in the primary dentition and more than six missing teeth in the permanent dentition. Hypodontia involving only maxillary primary canine was not reported in their study.

Studies have shown that agenesis of a primary tooth was often followed by agenesis of the permanent successor [3,4,7,12]. Daugaard-Jensen and co-workers found that agenesis of a primary incisor was followed by agenesis of the permanent successor in 86% of cases [12]. Nine primary second molars were found to be missing in their study and only one of them had a permanent successor. The cases in this report involved agenesis of only one or both maxillary canines in the primary dentition, and the corresponding successors were present in all three cases. One of them showed absence of three permanent second premolars, and the other two cases had all permanent teeth developing. A supernumerary tooth was even found in one of these two patients, which was an uncommon finding [13].

The higher incidence of hypodontia among firstdegree relatives of the affected individuals than in the general population suggested a significant genetic component in the aetiology, although environmental factors could not be ruled out [14]. More than 200 genes have been identified to have various roles in the control of tooth development [15,16]. Mutations of certain genes have been shown to result in oligodontia in human, and genetic factors leading to other types of hypodontia are still being searched [17]. Enucleation of primary canine tooth buds has been described in some Africa populations, and the affected children would present with clinically missing primary canines [18,19]. Nonetheless, this ritual custom is not known in Chinese culture and no such cases have been reported in ethnic Chinese. All cases in this report are boys, yet previous studies have found no significant sex difference in the prevalence of hypodontia in primary dentition [3-7]. It is not known if there is a male predilection of this type of hypodontia or it is just a coincidental finding.

In summary, the cases in this report are considered uncommon as hypodontia in the primary dentition involving only maxillary canines is rare. This article reports three such cases. In all of them, the corresponding successor was present but otherwise the permanent dentition showed diverse anomalies in tooth numbers, ranging from hypodontia to hyperdontia.

What this paper adds

- Hypodontia in the primary dentition involving only maxillary canines is rare.
- The permanent dentition of those affected showed diverse anomalies in tooth numbers, ranging from hypodontia to hyperdontia.

Why this paper is important to paediatric dentists

- Paediatric dentists may encounter children with congenitally missing primary canines in clinical practice.
- Early detection of developmental anomalies in the permanent dentition of these cases may allow early intervention.

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