International Journal of Paediatric Dentistry 2004; **14:** 446–450

Congenitally missing maxillary permanent canines: report of 32 cases from an ethnic Chinese population

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Summary. Cases of congenitally missing permanent teeth involving only maxillary canines are uncommon. Prevalence studies have revealed that it is a rare finding in Caucasian populations, but it may be relatively more common in Asian groups. This report describes radiographic findings in 32 Chinese children with congenitally missing maxillary permanent canines. The aetiology of such an anomaly is obscure, but the racial difference in prevalence suggests that genetic factors may be more influential than environmental ones.

Introduction

The prevalence of hypodontia varies according to the population studied [1]. Recent reports have shown that the prevalence of hypodontia in the permanent dentition (third molar excluded) is about 4·5–7·4% in Caucasians, and that the most commonly missing tooth is the mandibular second premolar [2,3]. In an earlier study, Davis found the prevalence of hypodontia in the permanent dentition amongst Chinese children to be 6·9%, but the most commonly missing tooth was the mandibular incisor, which suggested a racial difference in the pattern of hypodontia [4]. Congenitally missing permanent teeth involving only maxillary canines are rare, and most reports in the literature have included only a few cases [5–12]. Muller *et al*. studied the prevalence of hypodontia in 13 459 white American children and found only five such cases [13]. This report describes the radiographic findings in 32 Chinese children with congenitally missing permanent teeth involving only maxillary canines.

Case reports

Thirty-two cases of congenitally missing permanent teeth involving only maxillary canines were diagnosed at a school dental clinic during the 2001–2002 school year. The clinic was responsible for care for more than 70 000 primary school students in Hong Kong. Cases that involved other congenitally missing teeth (except third molars) were excluded from this report. Radiographs were taken only when anomalies or pathosis were suspected clinically. Indications for orthopantomograms included extensive lesions which could not be covered by intraoral radiographs, anomalies that involved several quadrants of the mouth, such as multiple missing teeth, patients who required multiple extractions, and patients whose canines were suspected to be missing or severely displaced. During the school year in which the study took place, 69 852 children attended the clinic for consultation, and a total of 1114 orthopantomograms were taken. (The total number of children examined was less than the total number of enrolled children since some did not attend for consultation during the year.) Orthopantomograms were taken for all cases included in this report, which ruled out the possibility of severe displacement that may not be seen with intraoral radiographs.

The details of the radiographic examination carried out are summarized in Table 1. A total of 19 girls and 13 boys were affected. All were ethnic Chinese. The age range was 6–14 years, with the majority of cases being diagnosed at the age of 11–12 years. Of the 32 cases, 11 were unilateral on the right side and 12 on the left. The remaining nine were bilateral. Five cases were associated with a microdontic

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Table 1. Radiographic findings in 32 cases of congenitally missing maxillary permanent canines: (M) male; and (F) female.

Case	Gender	Presenting age (years)	Missing teeth	Resorption of the corresponding primary predecessor	Other developmental dental anomalies (third molar excluded)
1	M	12	23	Little or no resorption	
2	F	7	23	Little or no resorption	
3	M	11	13	53 exfoliated	Microdontic 12 and 22 (barrel-shaped)
			23	63 exfoliated	
4	F	11	13	53 exfoliated	Microdontic 12 and 22 (peg-shaped)
			23	63 > 50% resorbed	
5	F	10	13	Little or no resorption	Microdontic 12 and 22 (barrel-shaped) (Fig. 1)
6	F	6	13	53 exfoliated	65 submerged, 41/42 double tooth (Fig. 2)
			23	63 > 50% resorbed	
7	M	12	13	53 < 50% resorbed	
8	F	11	23	63 < 50% resorbed	
9	F	10	13	53 exfoliated	
10	F	12	13, 23	Little or no resorption	
11	M	12	13	53 < 50% resorbed	
12	F	12	13	Little or no resorption	12 deep palatal pit
13	F	11	23	Little or no resorption	
14	F	11	23	63 exfoliated	
15	F	11	23	Little or no resorption	
16	M	11	13	53 > 50% resorbed	
17	M	12	13	53 > 50% resorbed	
18	M	12	23	63 > 50% resorbed	
19	F	12	13	53 > 50% resorbed	
			23	63 < 50% resorbed	
20	M	11	13	Little or no resorption	
21	F	11	23	Little or no resorption	Microdontic 13 (Fig. 3)
22	F	11	23	Little or no resorption	
23	M	11	13	53 < 50% resorbed	
			23	63 < 50% resorbed	
24	F	11	23	Little or no resorption	Microdontic 12 and 22 (peg-shaped)
25	F	11	13	53 < 50% resorbed	
26	F	12	23	63 > 50% resorbed	
27	F	13	23	63 exfoliated	47 mesially impacted (Fig. 4)
28	M	11	13	Little or no resorption	Microdontic 12 and 22 (peg-shaped)
29	M	14	13	53 exfoliated	
30	M	12	13, 23	Little or no resorption	
31	M	12	13, 23	Little or no resorption	
32	F	11	13	53 < 50% resorbed	
			23	63 > 50% resorbed	

maxillary lateral incisor (Fig. 1), one showed fusion of mandibular incisors (Fig. 2), the contralateral maxillary canine was microdontic in another (Fig. 3), and the mandibular permanent second molar was mesially impacted in one case (Fig. 4). The total number of congenitally missing maxillary permanent canines in the 32 cases was 41. Eight of the primary predecessors to the missing canines had already exfoliated and the predecessors of the remaining 33 were examined for degree of root resorption. Since accurate assessment of root length was not feasible with orthopantomograms, the degree of resorption was classified as little or no resorption, or less than half or more than half the root length resorbed. Based on this definition, 17 subjects showed little

Fig. 1. Case 5: Radiograph of case 5 showing a congenitally missing maxillary right permanent canine. Both maxillary permanent lateral incisors were barrel-shaped.

Fig. 2. Case 6: Radiograph showing that both maxillary permanent canines were congenitally missing. The mandibular right incisor was a double tooth and the maxillary left primary second molar was submerged.

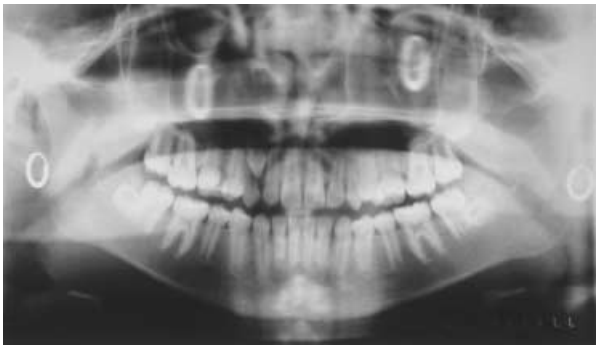

Fig. 3. Case 21: Radiograph showing a congenitally missing maxillary left permanent canine. The maxillary right permanent canine appeared to be microdontic.

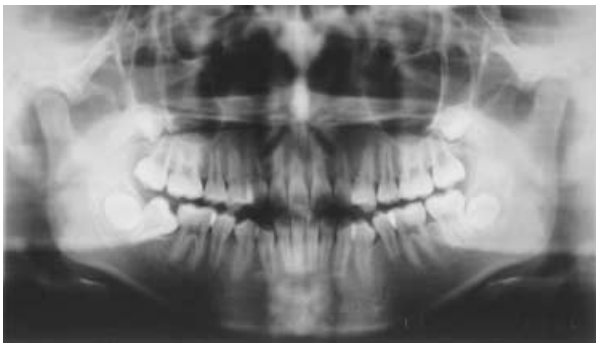

Fig. 4. Case 27: Radiograph showing a congenitally missing maxillary left permanent canine. The mandibular right permanent second molar was mesially impacted.

or no root resorption and the remainder presented with variable degrees of root resorption (see Table 1).

Discussion

The mechanism causing congenitally missing maxillary permanent canines in these patients is obscure. Butler's field theory states that the distal teeth within each morphological class are developmentally less stable [14]. According to this theory, the canine tooth stands alone in its own field, displaying great stability, and therefore, is rarely congenitally missing. Sofaer *et al*. suggested that the greater variability seen in teeth that form later in each morphological class may be a result of the interaction of tooth germs during development [15]. Bazan presented two cases of congenitally missing maxillary permanent canines associated with peg-shaped maxillary lateral incisors [7]. She suggested that, since maxillary permanent canines develop before lateral incisors, the absence of the canine might sufficiently alter the local environment at the time of development of nearby unstable teeth to result in a reduction in size. Brook proposed a multifactorial model incorporating genetic and environmental influences to explain anomalies of human tooth number and size [16,17]. His model involved a continuous scale with thresholds related to both tooth number and size. According to Brook's model, as tooth size is reduced, a threshold is crossed, at which point agenesis occurs. This model also explained the strong association between hypodontia and microdontia found in his study. Among the cases in the current report, the prevalence of microdontic maxillary lateral incisor was 16% (five of the 32 cases), which was higher than that of the local population, which had been found to be 3·3% [18]. This finding therefore concurs with the suggestions by Bazan [7] and Brook [16]. Such comparisons may not be entirely accurate, however, since the sample included in this report was small and selected.

The number of cases in this report is large when compared with previous case reports, mainly because of the large patient pool in the authors' clinic. Hallett and Weyman reported 14 cases of congenitally missing canines in 1954 [8], but only three of them met the criteria set in this report, i.e. only one or both maxillary permanent canines were congenitally missing. The number of cases included in this report cannot be regarded as representing the prevalence of congenitally missing maxillary canines in the local population, however, since the sample, although large, was limited to children attending a clinic and because radiographs were not routinely taken for all patients. Nevertheless, the large number seen suggests that racial difference may play a part. Muller *et al*. found only five cases

of congenitally missing permanent teeth involving only maxillary canines among 13 459 white American children (0·04%), whereas they found two such cases out of 1481 black children (0·14%) in the same study [13]. Davis also reported five such cases out of 1093 Chinese children in her study (0·46%) [4]. The racial differences seen in comparing these studies do seem to suggest that genetic factors may be influential in the aetiology of congenitally missing maxillary permanent canines. Markovic studied the pattern of hypodontia in twins and found a mirror-image pattern of unilaterally missing maxillary permanent canine in a pair of monozygotic twins [19]. This may give further support for a genetic basis, although the role of environmental factors cannot be ruled out.

Most cases in this report were diagnosed when the subjects were 11–12 years old, the age at which the maxillary permanent canines should be palpable clinically if they have still not erupted. All cases in this report were regularly attending patients of the School Dental Care Service, which was their sole dental care provider. A check of records ruled out any possibility of the missing canines having been extracted. Congenitally missing permanent canines have also been reported in patients with severe hypodontia or syndromes such as ectodermal dysplasia [20,21]. In this report, however, cases with other congenitally missing teeth in addition to missing canines were specifically excluded, and none of the cases presented any sign of systemic anomalies.

Haselden *et al*. showed that primary canines without permanent successors may survive for a reasonably long period and serve a useful function in patients with severe hypodontia [22]. It is not known if the same result can be applied to patients with mild hypodontia. In this report, over 40% of the maxillary primary canines without permanent successors showed little or no root resorption at the time of diagnosis.

Congenitally missing maxillary permanent canines pose a particular challenge in treatment planning. Factors to be considered include the condition of the primary predecessor, the number of missing teeth, the overall alignment and occlusion, and most importantly, the patient's and/or parents' preferences. Treatment options may include timely extraction of the primary predecessors to facilitate spontaneous space closure with or without further orthodontic alignment, or to keep the primary canines and replace them with a suitable restoration when they are lost. An advantage of retaining the primary predecessor is that, with the growing use of implants, alveolar resorption may be avoided until the late teens, providing the maximum potential for implant placement without the need for bone grafting. Each patient has to be assessed individually to decide on the most suitable treatment plan. Referral to an orthodontist and/or prosthodontist for definitive treatment will be needed for most cases.

Résumé. Les dents permanentes absentes congénitalement ne concernant que les canines est peu commun. Les études de prévalence révèlent que c'est un événement rare dans les populations caucasiennes, mais il est relativement plus fréquent dans les groupes asiatiques. Ce rapport décrit les données radiographiques recueillies chez 32 enfants chinois avec absence congénitale des canines maxillaires permanentes. L'étiologie d'une telle anomalie est obscure, mais les différences raciales de prévalence suggèrent que des facteurs génétiques peuvent avoir plus d'influence que les facteurs environnementaux.

Zusammenfassung. Angeborene Zahnzahlverminderung, welche nur die bleibenden Oberkiefer-Eckzähne betrifft, ist selten. Prävalenzstudien zeigen, dass dies in einer kaukasischen Bevölkerung ein seltener Befund ist, während es in einer asiatischen Population anscheinend häufiger zu beobachten ist. Die vorliegende Untersuchung beschreibt 32 röntgenologische Befunde von chinesischen Kindern Fehlen der bleibenden Oberkiefer-Eckzähne. Die Ätiologie dieser Anomalie bleibt unklar, aber die ethnischen Unterschiede der Prävalenz deuten auf einen genetischen Einfluss hin.

Resumen. La ausencia congénita de caninos permanentes que implica sólo a los caninos superiores es infrecuente. Estudios de prevalencia han revelado que es un raro hallazgo en poblaciones caucásicas, pero puede ser relativamente más común en grupos asiáticos. Este informe describe hallazgos radiográficos en 32 niños chinos con caninos permanentes congénitamente ausentes. La etiología de tal anomalía es oscura, pero la diferencia racial en la prevalencia sugiere que los factores genéticos pueden ser más influyentes que los factores ambientales.

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