

Bilateral Bimaxillary Bi-Rooted Primary Canines: Report of a Case

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Bilateral bi-rooted primary canines are a rare dental anomaly and the occurrence with a syndrome is even rarer. This dental anomaly has mostly occurred in African populations. This article reports a case of bi-rooted primary canines bilaterally located in both jaws of a 15-year-old Chinese boy who also suffered from cleidocranial dysostosis (CCD).

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INTRODUCTION

Reportedly, primary teeth exhibit fewer abnormalities with respect to size and shape than permanent teeth.¹ Pediatric dentistry and tooth morphology textbooks indicate that the normal root form of a primary canine is single in number and conical in shape.² Few primary teeth have additional roots and those that do, are usually primary molars. Bi-rooted primary canines are rare and only a few cases have been reported in the literature.³⁻⁷ The exact etiology is unknown but it has been postulated that it is related to an ingrowth of Hertwig's epithelial root sheath.⁸

Bi-rooted primary canines have been reported in Japanese, Afro-American and Caucasian subjects.⁸⁻¹⁴ The majority have occurred in African patients. To date there have been only ten cases of bi-rooted maxillary primary canines,^{3,4,9,15,11,13,8,14,16,17,25} and only three cases of this anomaly in both jaws.^{9,16,5} This case describes the occurrence of bi-lateral bi-maxillary bi-rooted canines in a Chinese male.

Case report

A 15 years old Chinese boy with a complaint of multiple missing maxillary and mandibular teeth was referred for specialist opinion. He was diagnosed with cleidocranial dysostosis (CCD). Family and social histories revealed that

the patient's mother had no features of CCD and because of the autosomal dominant mode of inheritance of CCD; the boy's father was invited by the Clinical Genetic Service for an assessment and counseling. The extra-oral examination of the boy revealed that he had a mesocephalic face; his chin was slightly deviated to the left side, he had an increased lower facial height. While intra-orally he was in the mixed dentition which was developmentally delayed and the teeth present in the oral cavity were 53, 62, 63, 65, 75, 72, 73, 82, 83, 84, 11, 16, 15, 14, 21, 24, 26, 36, 34, 31, 41, 45 and 46 (Figures 1a and 1b).

The radiographic examination revealed multiple impacted permanent teeth and supernumeraries (Figure 2). A



Figure 1a. The maxillary and mandibular arches showing the presence of normal primary and permanent teeth.

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Figure 1b. Frontal and lateral views showing the class III incisor relationship and primary canines with normal crown morphology.

CT scan was obtained for further investigation and evaluation of the teeth (Figure 3). Subsequently, the presence of two fused roots on the maxillary primary canines was confirmed after extraction (Figure 4a and b). The radiographs also revealed that the mandibular primary canines 73 and 83 had two roots and that the bifurcation of the roots was in the coronal third of both of the teeth. The mesio-distal width of 53, 63, 73 and 83 was 7 mm.

Multiple supernumeraries totaling seven in number were detected in relation to the following teeth (14, 22, 32, 34, 35, 42, and 43). Examination of the remainder of the dentition



Figure 3. CT scan showing the bi-lateral, bi-maxillary bi-rooted primary canines and multiple impacted permanent teeth and supernumerary teeth in relation to 14, 22, 32, 34, 35, 42, and 43.

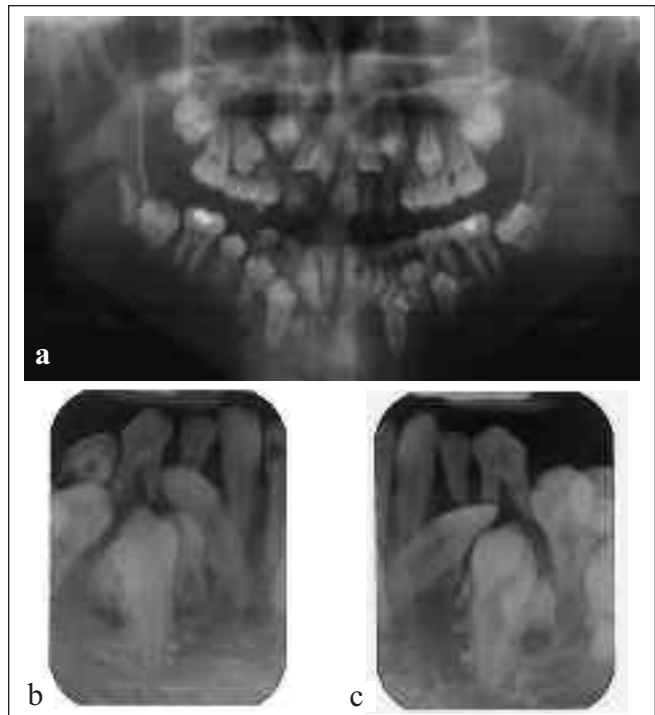


Figure 2. Panoramic radiograph showing multiple impacted supernumerary teeth in relation to 14, 22, 32, 34, 35, 42, and 43. The periapical radiographs showing the presence of bi-rooted primary canine 73 (b) on the right and (c) on the left side of the mandible.

revealed that the maxillary arch was short but symmetrical; clinically 11 was missing while 21 was erupting. The mandibular arch was also short and symmetrical; teeth 31 and 41 were rotated and retroclined. The molars on the left side were in a class III relationship, while the molars on the right side were in a class I relationship (Figure 1b). The patient also had a reverse overjet (Figure 1b). As part of the treatment plan it was decided to extract the primary teeth that were present to allow the adjacent impacted permanent teeth to erupt.

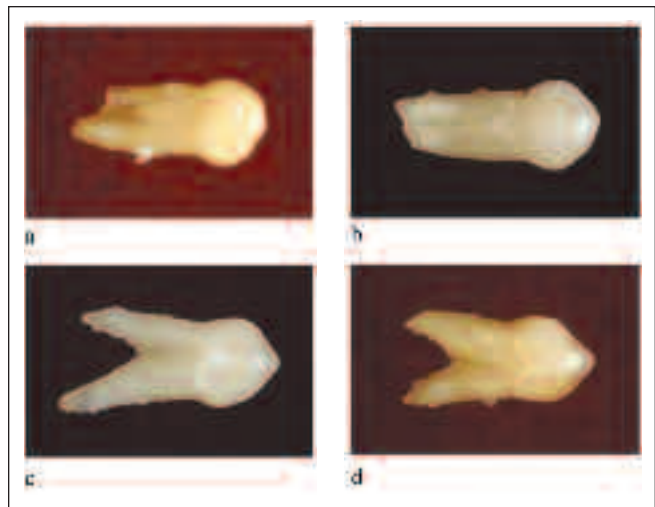


Figure 4. The buccal views of (a) maxillary right, (b) maxillary left, (c) mandibular right and (d) mandibular left canines.

DISCUSSION

The prevalence of bi-rooted primary canines appears to be higher in the maxilla than in the mandible. Of the 17 cases

of bi-rooted primary canines that were found in the literature, 14 cases were confined to a bilateral occurrence in the maxilla; while 3 cases involved both the maxilla and mandible bilaterally,⁶ see Table 1. With respect to the racial predilection of the occurrence of bi-rooted bilateral primary canines, 13 of the cases were observed in Afro-American populations indicating the anomaly is more common in black populations. The prevalence of bilateral bi-rooted primary canines has so far been reported in normal healthy individuals. The reported case is the first Chinese subject, with an associated syndrome and bilateral bi-rooted mandibular and maxillary primary canines to be reported in the literature.

The etiology of teeth with supernumerary roots is poorly understood. It has been postulated⁸ that bifurcated roots might be related to ingrowths of Hertwig's epithelial root sheath. Other researchers have suggested that fusion or ger-

Table 1. Showing the mesio-distal widths (mm) of primary teeth.

Tooth (primary)	Yuen, 1995		Otani <i>et al.</i> 1993	
	M	F	M	F
Mx central incisors	6.682	6.678	6.77	6.55
Mx lateral incisors	5.461	5.445	5.33	5.53
Mx canines	6.742	6.681	6.46	6.66
Mx first molars	7.406	7.263	7.35	7.23
Mx second molars	9.256	9.162	9.28	9.17
Mn central incisors	4.168	4.214	4.34	4.10
Mn lateral incisors	4.667	4.705	4.80	4.76
Mn canines	5.900	5.881	5.92	5.80
Mn first molars	8.184	8.095	8.26	8.06
Mn second molars	10.295	10.146	10.38	9.93

Table 2. Previously reported cases of bi-rooted canines adapted from Mochizuki⁵; Orhan & Sari.⁶

Author/year	Canine	Location	Age (yr)	Sex	Race	Siblings	Syndrome
Takano 1941	mn	unilateral	9	m	Jap	?	no
Kurosu <i>et al.</i> 1968.							
Case 1	mx	unilateral	8	f	Jap	?	no
Case 2	mx	unilateral	8	m	Jap	?	no
Case 3	mn	unilateral	8	m	Jap	?	no
Yasunaga <i>et al.</i> 1978	mx mn	bilateral	6	m	Jap	?	no
Hata <i>et al.</i> 1979	mn	bilateral	4	m	Jap	?	no
Brown 1975	mx	bilateral	4	f	?	?	no
Kelly 1978	mx	bilateral	5	f	black	?	no
Krolls & Donahue 1980	mx	bilateral	5	f	black	?	no
Chow 1980	mx	bilateral	5	m	black	?	no
Bimstein & Bystrom 1982	mx	bilateral	5	m	black	sister (13 yr): NAD	no
Bryant & Bowers 1982	mx mn	bilateral	5	m	Cauc	two older sisters: NAD	no
Paulson <i>et al.</i> 1985	mx	bilateral	9.6	m	black	?	no
Jones & Hazelrigg 1987	mx	bilateral	5	m	black	brother (10 yr): NAD	no
Saravia 1991							
Case 1	mx	bilateral	4.6	m	black	?	no
Case 2	mx	bilateral	4	m	black	?	no
Hayutin & Ralstrom 1992							
Case 1	mx	bilateral	4	m	black	?	no
Case 2	mx	bilateral	1.8	m	black	?	no
Ott & Ball 1996							
Case 1	mx	bilateral	8.6	m	black	older sister: NAD	no
Case 2	mx	bilateral	4.11	m	black	older sister: NAD	no
Case 3	mx mn	bilateral	8.4	m	black	no siblings	no
Winkler & Ahmad 1997	mx	?	4	f	Pueblo	no siblings	no
Mochizuki <i>et al.</i> 2001	mx mn	bilateral	6.4	m	Jap	?	no
Orhan & Sari 2006							
Case 1	mx	unilateral (63)	11.3	m	Cauc	younger sister (9 yr): NAD	no
Case 2	mx	unilateral (53)	4.9	m	Cauc	no siblings	no
Case 3	mx	bilateral	6	m	Cauc	no siblings	no

mination maybe related to the clinical presentation of the supernumerary roots.^{18-21, 24}

In this case the mesio-distal width the crowns of all the primary canines was 7 mm, which is larger than 5.9 mm, which has been reported for the primary canines in male Hong Kong Chinese.^{22, 23}

There is no reason why a bi-rooted primary canine should be extracted. It can be expected to function and exfoliate normally unless space management is required in a patient with a developing malocclusion. The bi-rooted primary canines were extracted in this case to permit the eruption of the underlying impacted permanent teeth. The extracted primary canines are shown in Figure 4. The detailed treatment planning of the problems related to CCD and the outcome will be discussed in a future paper. This case serves as a reminder of the importance of taking a radiograph even for the routine extraction of a primary canine.

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