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Case Report

First report of bilateral supernumerary teeth associated with both primary and permanent maxillary canines

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Abstract: A very rare case of bilateral supernumerary teeth in the maxillary canine region and its management through a 3-step-conservative approach and 5-year-follow up is presented in this report. A 7-year-old Caucasian boy presented with 2 erupted supernumerary primary maxillary canines (53s and 63s) and two unerupted supernumerary permanent maxillary canines (13s and 23s). The treatment was carried out in three steps. In the first step, we removed teeth 53s and 63s. As the second step, early removal of teeth 53 and 63 and cementation of a space maintenance appliance (Nance's arch) were performed. In the third step, teeth 13s and 23s were removed, and the Nance's arch was maintained until the complete eruption of teeth 13 and 23. The management of this case with a proper treatment plan enabled us to solve the problem without complex procedures. (J Oral Sci 51, 145-150, 2009)

Keywords: tooth, supernumerary; diagnosis; treatment outcome.

Introduction

The causes of supernumerary teeth are poorly understood, although many theories have already been proposed, such as the phylogenetic process of atavism and the dichotomy of the tooth bud. The most accepted theory suggests that these teeth result from localized and independent hyperactivity of the dental lamina, which presumably leads to the formation of additional tooth germs (1-5). Genetics is also thought to play a role in the development of supernumerary teeth, as recurrence within the same family is commonly reported (6,7). The occurrence of similarly located supernumerary teeth in monozygotic twins also suggests the influence of genetic factors (8). In addition, it is believed that environmental factors along with hereditary factors are combined to cause the condition (9).

Numerous hereditary syndromes have been described in association with hyperdontia, such as Cleidocranial dysplasia, Crouzon syndrome, Ehlers-Danlos syndrome, Gardner syndrome, Nance-Horan syndrome, and cleft lip and palate (3). Mutations in the genes *RUNX2*, *APC* and *NHS* are associated with the etiology of Cleidocranial dysplasia, Gardner and Nance-Horan syndromes, respectively (10). However, no mutations providing non-syndromic supernumerary teeth have been identified yet (11).

Supernumerary teeth are positively correlated with macrodontia (3). Studies have shown that the teeth of individuals with hyperdontia tend to be larger than average, especially the mesiodistal dimension of the crown region. These findings suggest that anomalies of tooth number and size may have similar etiologies (12,13).

Although supernumerary teeth may occur either unassociated to any other anomaly or as part of developmental disorders, multiple supernumerary teeth are rare in individuals with no other associated diseases or syndromes (4,5). They can have rudimentary morphology or normal size and shape, the latter being called supplemental (1,2,4,5). They are more frequently seen in

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the maxillary region of the permanent dentition, especially the mesiodens, which is located in the premaxilla (3-5,14-18). The frequency of occurrence, in decreasing order, is mesiodens, maxillary fourth molars, maxillary paramolars, mandibular premolars, and maxillary lateral incisors (3,5,17). When they affect the primary dentition, they usually appear as maxillary lateral incisors, while mesiodens, maxillary canines and mandibular incisors have also been reported (14,17,19-21). A sex predominance of males over females has also been reported (4,9,14,15,18,22).

The presence of supernumerary teeth may give rise to a variety of clinical problems, such as dentigerous cyst (23), malocclusion (23-25), root resorption (1,5,18), displacement or rotation (2,18,25), failure of eruption or delayed eruption of the adjacent tooth (2,18,23,26), esthetic disturbances (25), and others (1,5,15,18). Findings from surveys have shown that most supernumerary permanent teeth remain unerupted (4,18). Additionaly, as these teeth can be asymptomatic, a thorough assessment of X-rays covering all the teeth and adjacent structures and the use of the parallax radiographic principle are recommended in order to increase diagnosis accuracy (4,15,16,18).

World-wide, many studies have reported the prevalence of supernumerary canines, which has been estimated from 0 to 1.5% (4,14-16,18-20,22). In a study involving 1,700 American adolescents, 64 supernumerary teeth distributed across 39 individuals were detected; no supernumerary canines were reported (22). Miyoshi et al. (19) evaluated the prevalence of supernumerary primary teeth in 8,122 Japanese children aged 3 to 6 years old. There were only 4 cases (0.05%) of supernumerary primary teeth and all of them were located in the maxillary lateral incisor area. A study of occurrence of dental anomalies in the primary dentition of Malaysian children detected that 4 subjects out of 65 had supernumerary teeth. Of these, 3 were mesiodens and 1 was a mandibular lateral incisor (20). Among 152 Jordanian children aged 5 to 15 years old, the prevalence was 1.5% and 1% of supernumerary maxillary and mandibular canines, respectively (4). In a survey of the frequency of supernumerary teeth in a Mexican population, Salcido-García et al. (16) studied the radiographs of 2,241 patients with a mean age of 14 years. The prevalence of supernumerary teeth was 3.2%, and only 1 patient had a supernumerary canine. Likewise, Primo et al. (15) evaluated the presence of supernumerary teeth in 2- to 14-year-old Brazilian children. Seventy supernumerary teeth were found among 1,907 dental record forms; there was only 1 canine (1.4%). In another Brazilian study, which assessed the demographic profile of 460 supernumerary teeth in children and adolescents aged 3 to 16 years old, the prevalence of maxillary canines was 2.6% and that of mandibular canines was 1.3% (18). Finally, a survey on the prevalence of dental anomalies among 1,878 Saudi Arabian children detected 11 children with supernumerary primary teeth and 24 with supernumerary permanent teeth; the relative frequency of canines was 10% (14).

As far as we are aware, a non-syndrome case with bilateral supplementary primary and permanent maxillary canines has not yet been reported. Thus, in this report, we present a very rare case of bilateral supernumerary teeth associated with both primary and permanent maxillary canines, and describe a conservative approach involving 3 steps completed over a period of 5 years.

Case report

A 7-year-old Caucasian boy visited the Pediatric Dentistry Clinic of the School of Dentistry, Rio de Janeiro State University, in order to undergo a routine dental examination. A medical history was taken, and clinical and radiographic examinations were carried out. According to the medical history, neither systemic disorders nor hereditary patterns of hyperdontia in the family were detected. Likewise, there was no symptom suggestive of a syndrome background.

Intra and extra oral clinical examinations did not reveal any disturbances in soft or hard tissues. Clinical examination revealed that the patient was in the mixed dentition phase with the presence of the following teeth: 16, 26, 36, 46, 31, 32, 41, 42, 11, 21, 53, 54, 55, 63, 64, 65, 73, 74, 75, 83, 84, and 85. Apart from these teeth, there were also two supernumerary primary maxillary canines (53s and 63s), which were located mesial to teeth 53 and 63. Teeth 53 and 53s and 63 and 63s were practically identical and in good alignment. No maxillary lateral incisors were present, but the patient's mother confirmed that both 52 and 62 had shed normally (Fig. 1). The occlusal examination showed that the patient was Angle's class I and had normal overbite and overjet. A panoramic radiograph demonstrated the presence of 2 erupted primary canines in each quadrant of the maxilla (53, 53s, 63, 63s) and the 4 unerupted permanent canines (13, 13s, 23, 23s – Fig. 2). The same radiograph revealed that teeth 12 and 22 were impacted due to the roots of teeth 53s and 63s.

After confirming the diagnosis of hyperdontia in the maxillary canine region, the treatment was carried out in three steps. The first step included oral hygiene and dietary counseling, followed by removal of 53s and 63s in order to allow the eruption of 12 and 22 (Fig. 3). In the second step, 53 and 63 were extracted so that teeth 13s and 23s could erupt and a space maintenance appliance (Nance's

arch) could be cemented (Figs. 4 and 5). The third step consisted of removal of teeth 13s and 23s and maintenance of the Nance's arch (Fig. 6). After the eruption of teeth 13 and 23 at the age of 12, the Nance's arch was removed and the complete occlusion was established (Figs. 7 and 8).

Discussion

There is a consensus regarding the localization of the supernumerary teeth; most of them occur in the maxilla and in the permanent dentition, the mesiodens being the most common (3-5,15,16,18). According to these authors, the canine is rarely affected.

As previously mentioned, we failed to find literature on bilateral supernumerary primary and permanent maxillary canines. In the case described by Hume (27), a 17-year-old boy was found to have 4 supernumerary permanent canines, 2 in the maxilla and 2 in the mandible. Cruz and Campos (24) reported the occurrence of supernumerary maxillary right canines in both dentitions (53s and 13s) in a 9-year-old girl. Türkkahraman et al. (25) described a rare case of a 12-year-old boy, who did not suffer from any systemic disease or syndrome, presenting bilateral supernumerary permanent maxillary canines (13s and 23s) of normal shape and size. Shetty and Sandler (23) described a case of 2 supernumerary permanent teeth in an 8-year-old boy: 1 palatal to the crown of the maxillary left central incisor and the other a maxillary left canine.

As for the primary dentition, Ismail (26) described the presence of 2 supernumerary primary maxillary canines (53s and 63s) in a 10-year-old boy, whereas Hayduk and Stout (2) reported 2 supernumerary primary maxillary canines (53s and 63s) as well as a supernumerary permanent maxillary right canine (13s) in an 8-year-old girl. Ferguson and Hauk (28) showed a case of a 4-year-old boy presenting

Fig. 1 Frontal view on first clinical examination.

Fig. 3 Eruption of teeth 12 and 22 after the extraction of teeth 53s and 63s.

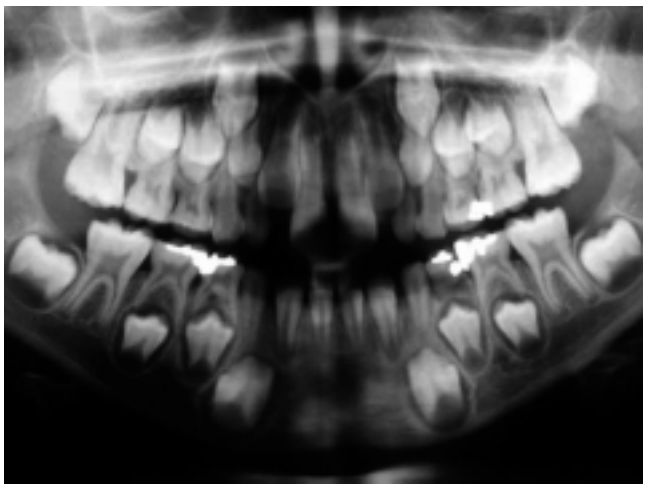

Fig. 2 First panoramic radiograph showing four supernumerary canines.

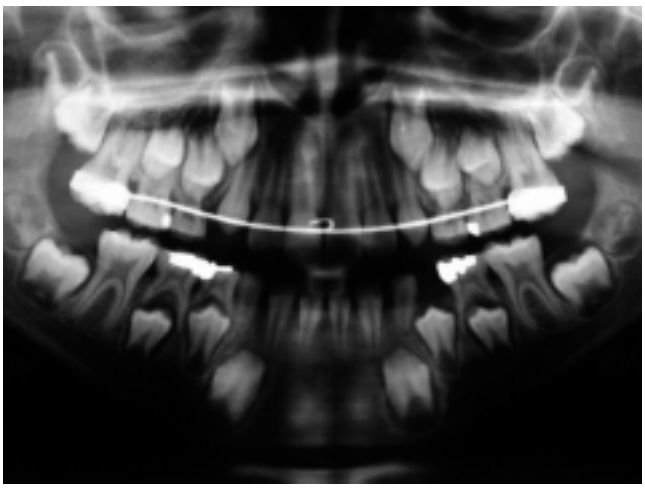

Fig. 4 Radiograph after the early removal of teeth 53 and 63 and cementation of a Nance arch.

a single supernumerary primary maxillary left canine (63s). Roberts et al. (21) reported a case of a healthy 22-month-old boy whose intraoral examination revealed the presence of a supplementary 51 and bilateral supplementary primary maxillary canines (53s and 63s).

Different approaches to deal with such conditions have been reported in the literature. The authors agree that an early diagnosis is of paramount importance to minimize the risks of complications resulting from supernumerary teeth. However, the best time for removal of a supernumerary tooth depends on careful evaluation of each situation (1,4). Some authors suggest immediate removal of the tooth so as to prevent costly future orthodontic intervention (2); the others claim that the extraction of asymptomatic supernumerary teeth that do not affect the dentition may not always be necessary, although they should be periodically monitored (4,21). Furthermore, when supernumerary primary teeth are detected, parents should be warned of the possible consequences to the permanent dentition as these teeth may be replicated in the permanent series in 50% of the cases (20).

The cases mentioned in this paper reflect the numerous ways to manage such dental anomalies. Ismail (26) decided to extract the teeth 53s and 63s in order to provide more space for the erupting 12 and 22. Hayduk and Stout (2) also employed extraction of both 53s and 63s and for tooth 13s. Cruz and Campos (24), after clinical and radiographic examination, noted that the supernumerary teeth were affecting the occlusion. Therefore, it was decided to perform simultaneous extraction of teeth 53s

Fig. 5 Eruption of teeth 13s and 23s after the extraction of teeth 53 and 63.

Fig. 7 Radiograph after the eruption of teeth 13 and 23 and removal of the Nance arch.

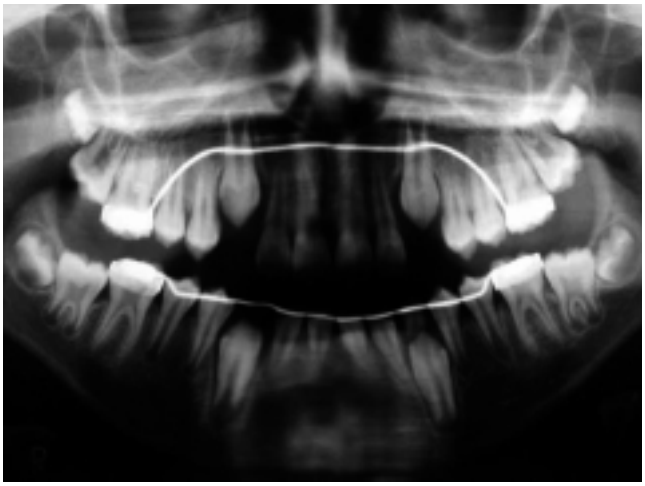

Fig. 6 Radiograph after the removal of teeth 13s and 23s and maintenance of the Nance arch.

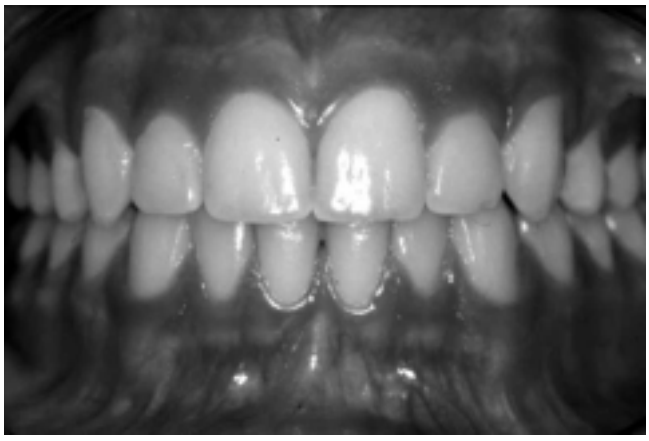

Fig. 8 Frontal view after the treatment at the age of 12.

and 13s. On the other hand, Roberts et al. (21) and Ferguson and Hauk (28) advocated monitoring without removal. Shetty and Sandler (23) recommend the extraction of the supernumerary palatal to tooth 21, but decided that removal of the canine could be harmful at that time. A dentigerous cyst was detected 3 years later and the patient had the cyst enucleated under general anesthesia.

As the majority of supernumerary teeth lead to clinical complications (3), the standard of care is early removal. However, the importance of case-by-case assessment should be highlighted. Some factors can have a significant contribution to the failure of spontaneous eruption of an impacted tooth after removal of a supernumerary tooth: supernumerary tooth with a tuberculate form, presence of multiple supernumerary teeth, time of surgery occurring after estimated time of eruption of the impacted tooth, early stages of root formation of the supernumerary tooth, and space loss. Thus, under these unfavorable conditions, prompt orthodontic traction is recommended concomitantly with the removal of the supernumerary tooth (29,30).

In the present case, extraction of teeth 53s and 63s allowed the teeth 12 and 22 to emerge spontaneously in 30 days and to maintain a favorable position in the arch (Fig. 3). It was also decided on early removal of teeth 53 and 63 to allow the eruption of teeth 13s and 23s (Fig. 5). This approach was successful and a simple extraction without complex procedures such as flap and osteotomy was performed after the eruption of teeth 13s and 23s. The space was maintained by placement of the Nance's arch until complete eruption of teeth 13 and 23 at the age of 12.

Based on the literature and the case described herein, it is possible to conclude that early diagnosis and treatment enabled the child and his family to solve the problem without traumatic surgical procedures or corrective orthodontic treatment. We believe that this report would help other professionals to deal with similar cases.

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