Multiple bilateral supernumerary mandibular premolars in a non-syndromic patient with associated orthokeratised odontogenic cyst- A case report and review of literature

Vikrant O. Kasat, Harish Saluja[1], Jitendra V. Kalburge[2], Yogesh Kini[1], Atul Nikam[2], Ruchi Laddha[3]

Abstract

Multiple supernumerary teeth are very rare, accounting for less than 1% of cases. They are commonly associated with syndromes like Gardner's syndrome and cleidocranial dysostosis and cleft lip and palate. Non-syndromic multiple supernumerary teeth have a predilection to occur in the mandibular premolar region. Orthokeratinized odontogenic cyst (OOC) is a relatively uncommon developmental cyst comprising about 10% of the cases that had been previously implied as odontogenic keratocysts. More than half of the cases of OOC are associated with impacted tooth; but not a single case of OOC associated with supernumerary teeth is reported. Hence, the purpose of this article is to report the first case of multiple supernumerary mandibular premolars associated with OOC in a 35-year-old male and to review the literature associated with multiple bilateral supernumerary mandibular premolars.

Keywords: Impacted, mandibular, orthokeratinized odontogenic cyst, supernumerary premolars

Introduction

Supernumerary teeth or hyperdontia is a mammalian developmental abnormality characterized by the presence of extra teeth in addition to teeth of the normal eruption series.[1] These teeth may be single or multiple, unilateral or bilateral, erupted or impacted, and in one or both jaws.[2] The prevalence of supernumerary teeth varies between 0.3% to 1.9% in primary dentition and 0.1% to 3.6% in permanent dentition.[1] Even though the majority of supernumerary teeth appear in the premaxillary region (mesiodens), supernumerary premolars (SP) have a predisposition for the mandible.[3] It has been reported that the prevalence of the SP is between 0.029[4] - 0.64%,[5] and premolars account for only 10% of all the supernumerary cases.[4] Single supernumeraries occur in 76-86% of cases, double supernumeraries occur in 12-23% of the cases, and multiple supernumerary teeth are very rare, accounting for less than 1% of cases.[6]

Departments of Oral Medicine and Radiology, [1]Oral and Maxillofacial Surgery, [2]Oral Pathology and Microbiology and [3]Prosthodontics, Rural Dental College, Loni, Maharashtra, India

Correspondence: Dr. Vikrant Kasat, Department of Oral Medicine and Radiology, Rural Dental College, Loni- 413 736, Maharashtra, India. E-mail: drvikrantkasat@rediffmail.com

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Orthokeratinized odontogenic cyst (OOC) is a relatively uncommon developmental cyst comprising 5.2% to 16.8% of cases that had been previously implied as odontogenic keratocysts (keratocystic odontogenic tumor). In 1981, Wright specified its clinicopathological aspects and suggested that it be called "odontogenic keratocyst- orthokeratinized variant." The term "orthokeratinized odontogenic cyst" was suggested by Li.[7]

Dental literature is replete with cases of supernumerary mandibular premolars and orthokeratinised odontogenic cysts as separate entities; but to our knowledge, the present clinical scenario has not been reported previously. The purpose of this article is to report the first case of MSMP associated with OOC in a patient who had multiple supernumerary mandibular premolars bilaterally and to review the literature associated with MBSMP. A search of "PubMed" was made with the keywords "bilateral mandibular supernumerary premolars," "multiple supernumerary teeth," "orthokeratinised odontogenic cyst associated with impacted tooth," and "orthokeratinised odontogenic cyst associated with supernumerary tooth." It was supplemented with hand search to identify related published articles in dental journals. For review, only those articles were selected, in which MBSMP were present (minimum two SP on each side). From review, it is clear that non-syndromic MBSMP are rare, and as far as we could determine, only 16 cases have been reported since 1985 till date. [Table 1]

Case Report

A 35-year-old male reported to the Dept. of Oral Medicine and Radiology with the chief complaint of pus discharge from lower left posterior region of jaw since one week. Detailed

Table 1: Summary of case reports of multiple bilateral supernumerary mandibular premolars (Minimum 2 on each side) in chronological order

First Author	Year	Age	Sex	Mandible		Associated SP in maxilla	Status of mandibular SP	Associated pathology
				R	L			
Kantor[6]	1988	9	M	2	2	4	Unerupted	None
Rubenstein[5]	1991	13	F	2	2	-	Unerupted	None
Hopcraft[3]	1998	18	M	3	2	3	Unerupted	None
Saini[1]	2002	19	F	2	2	2	Unerupted	None
Farahani[2]	2007	13	F	2	3	-	Unerupted	None
Hyun[4]	2008	11	M	2	2	-	Unerupted	None
		13	M	2	2	-	Unerupted	None
		20	F	3	2	-	Unerupted	None
		13	M	2	2	-	Unerupted	None
		16	M	2	2	-	Unerupted	None
		17	M	2	3	-	Unerupted	None
		11	M	2	2	-	Unerupted	None
		13	F	2	2	-	Unerupted	None
		28	M	2	2	-	Unerupted	None
Bhatia[8]	2010	12	F	2	2	-	Unerupted	None
Kaya[9]	2011	39	F	2	2	1	Unerupted	None
Current case	2011	35	M	2	2	-	Unerupted on left side and erupted on right side	Orthokeratised odontogenic cyst on left side

SP = Supernumerary premolars

history revealed that it started as swelling 2 months back, for which he took medicines for 2 weeks. But no permanent relief was obtained, and condition further deteriorated when pus discharge from left posterior region of jaw started 1 week back. Later on, lower left first molar was extracted for the same reason 3 days back. As there was no relief, patient reported to our dental college.

Extraoral examination revealed presence of a diffuse, firm to hard, tender swelling over left body of mandible. Intraoral examination revealed extraction socket of the lower left first molar. There was expansion of buccal and lingual cortical plates and loss of vestibular depth from left lateral incisor to first molar region. Left second premolar had grade II mobility. Teeth from right lateral incisor to left second premolar were tender on percussion. Also, two SP were seen on lingual aspect of lower right first and second premolars [Figure 1]. A provisional diagnosis of cystic or benign lesion was considered.

Radiographic evaluation included intraoral periapical radiograph [IOPA] of the lower left first molar, mandibular occlusal view, and orthopantomograph [OPG]. It revealed a well-defined corticated radiolucency containing two supernumerary teeth. It was extending anteroposteriorly from distal aspect of lower right canine till distal aspect of left second molar. Superiorly, it was extending up to the alveolar ridge and inferiorly up to the inferior border of mandible causing thinning of the inferior border [Figure 2]. Expansion of buccal and lingual cortical plates was seen [Figure 3]. Differential diagnosis of dentigerous cyst, keratocystic odontogenic tumor, and unicystic ameloblastoma was considered.

Incisional biopsy report was indicative of OOC. Patient was referred to maxillofacial surgery department where enucleation of cystic lesion was carried out under general anesthesia after the root canal treatment of involved teeth was completed. The lesion was enucleated *in toto* along with the two impacted SP [Figure 4]. Peripheral osteotectomy was done. The enucleated lesion was sent for histopathological examination, which confirmed the finding of incisional biopsy report [Figure 5]. Postoperatively, the surgical site was covered with an acrylic plate to facilitate healing [Figure 6]. The healing was uneventful, and patient is being followed up for last 8 months and is asymptomatic [Figure 7].

Discussion

According to the literature, only 2% of the SP are likely to undergo pathological changes. Though supernumerary teeth may cause various clinical problems, dentigerous cyst

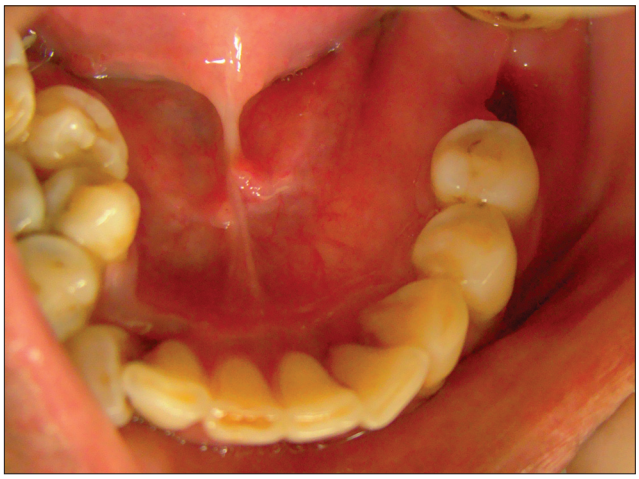

Figure 1: Intraoral photograph showing swelling on left side and two supernumerary mandibular premolars on right side

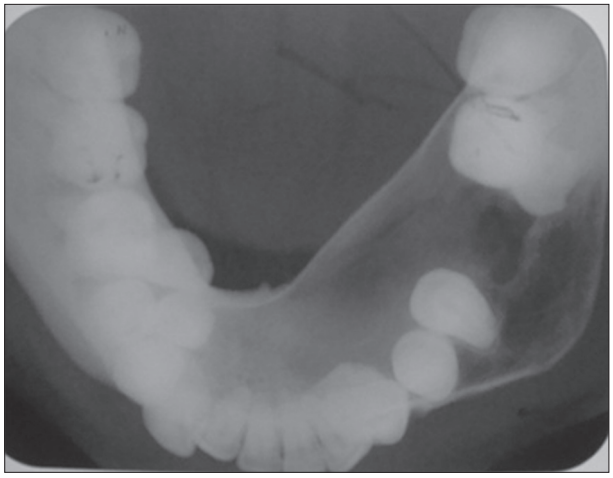

Figure 3: Mandibular occlusal view revealing expansion of buccal and lingual cortical plates on left side and two supernumerary premolars on right side

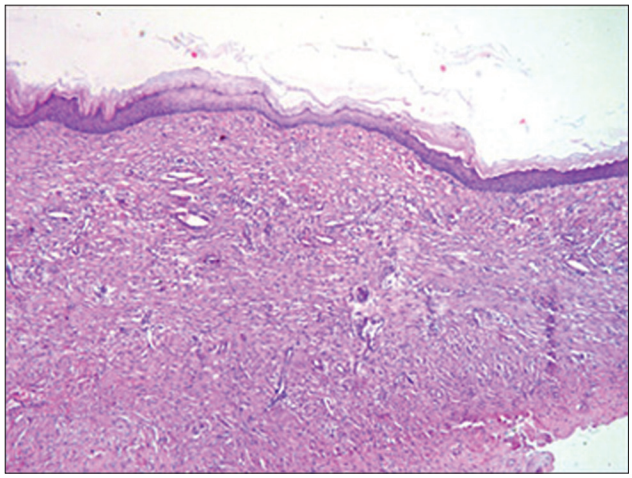

Figure 5: Photomicrograph shows hyperorthokeratinized stratified squamous epithelium with surface corrugations and the fibrocellular connective tissue. (H and E Stain, ×10)

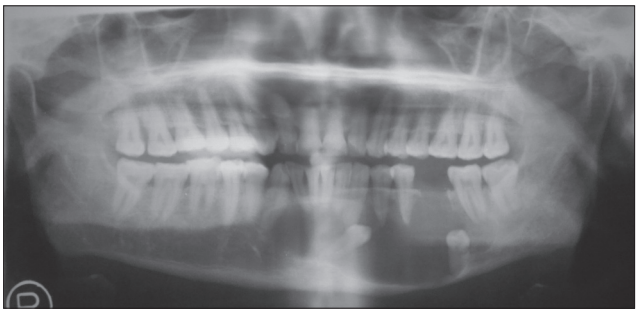

Figure 2: OPG showing a well defined corticated radiolucency containing two supernumerary premolars

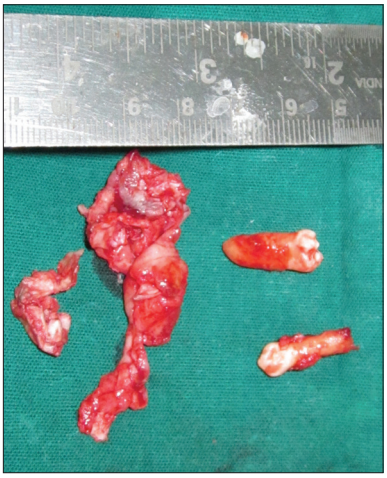

Figure 4: Excised lesion with two supernumerary premolars

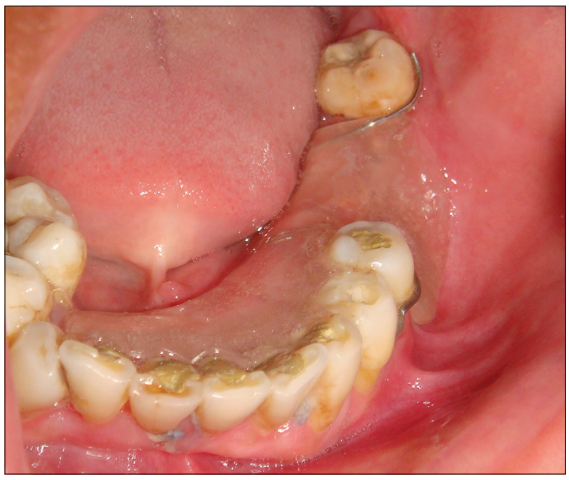

Figure 6: Acrylic plate covering the surgical site

formation and root resorption have been cited in the literature as frequent complications associated with SP.[9] In all the cases of MBSMP reviewed, none was associated with any pathology. Ours is the first report of MSMP associated with OOC.

Several factors might explain the apparent discrepancy in the prevalence figures of MSMP reported, such as

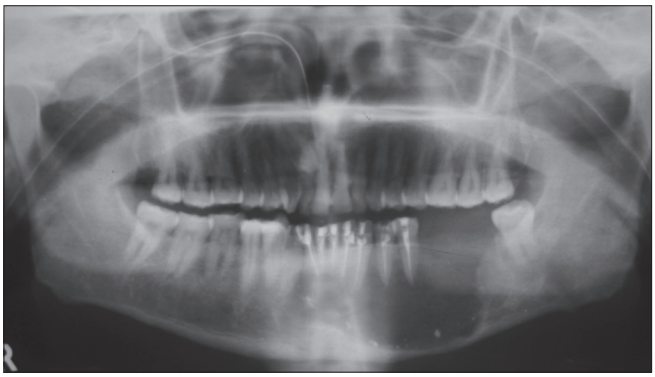

Figure 7: Post-operative OPG

differences in patient population samples, age groups, ethnicity, radiographic techniques used, and the possibility that supernumerary teeth may have been extracted before examination.[5]

Even though the exact etiology of supernumerary teeth is not known, several hypotheses have been proposed. These include phylogenetic theory of atavism (evolutionary throwback), dichotomy theory (cleavage of a single tooth bud to two homologous or heterologous parts),[2] morphogenetic field theory,[1] and heredity (an autosomal dominant trait,[8] sex-linked inheritance.[10]) The most widely accepted hypothesis is the hyperactivity theory suggesting supernumeraries are the result of localized, independent, and conditional hyperactivity of dental lamina. The symmetrical bilateral development in our case as well as in reviewed literature indicates that other factors are involved in the development of supernumerary teeth in addition to localized hyperactivity of dental lamina. Thus, the etiology of supernumerary teeth appears to be multifactorial, being a combination of environmental and genetic factors.

The age in the cases reviewed ranged from 9 to 39 years with average of 16.5 years. Among the cases reviewed, majority occurred (14/16) in patients under the age of 20 years.[1-6,8] One patient each was in third[4] and fourth decade.[9] [Table 1] According to the literature, the SP are more frequent in males than in females,[4] but occasionally female predominance is mentioned (male to female ratio 1:2.3).[9] Among the 16 case reports summarized in Table 1, the incidence is higher in men ($n = 10$) compared with women ($n = 6$) with male to female ratio of 1.66:1. OOC occur in the third to fourth decade with an average age of 38.9 years. OOC has a male predominance with a male to female ratio of 2.59:1.[7] In our case, patient was a male in fourth decade. The finding that most of the cases reviewed were diagnosed and treated earlier (second decade) and OOC usually occur in fourth decade could explain partially why no pathology was found in reported cases.

The most common location of OOC is the mandibular molar and ramus region,[7] and the most common location of non-syndromic multiple supernumery teeth is mandibular premolar area.[11] Both of these findings were observed in our patient. According to morphology, supernumery teeth may be categorized as: Conical (a small peg-shaped tooth), tuberculate (barrel-shaped with incomplete root formation and often paired), supplemental (morphology similar to a tooth of the normal dentition), or odontoma (hamartomatous malformation).[2] In most cases, SP tend to be supplemental.[3] In the present case also, supernumeraries are of the supplemental type.

Three types of supernumerary teeth are described in the literature depending on the time of abnormal proliferation of the dental lamina i.e. predeciduous type, pre-permanent type (before development of the permanent tooth - more frequent type), and post-permanent type (after the deciduous as well as the permanent follicles have been formed). In our case, they belonged to pre-permanent type.

The majority of SP do not erupt into the oral cavity. In all the cases reviewed, SP were unerupted i.e. either impacted or developing. The present case differed in that SP on right side were erupted in the oral cavity and were placed lingually.

Since a large percentage of SP remain impacted, unerupted and are generally asymptomatic, radiographs play an important role in diagnosis. For early radiographic diagnosis of SP, panoramic examination can be done from 12 to 14 years of age and can be repeated at 5-year intervals. Out of 16 cases of MBSMP reviewed, 4 had associated SP in maxilla.[1,3,6,9] Thus, importance of panoramic examination cannot be overlooked.

In the reviewed literature, recurrence of SP after being surgically removed has been reported.[5] These authors did not give a possible explanation as to why these teeth may have recurred. It is possible that the crypts of SP could have been present earlier, but were not detected in the previous radiographs. Another possibility is that the dental lamina is not resorbed completely and is reactivated at the time of crown completion of normal permanent teeth. Incomplete resorption of the dental lamina creates multiple supernumerary teeth, especially in the premolar region.[10]

Radiographically, OOCs more frequently present as unilocular radiolucencies (87.0%), and about 60.8% of the cases reported in the literature were found to be associated with an impacted tooth.[7] Though in our case it presented as unilocular radiolucency, it differed from reported cases in literature in that it was associated with multiple impacted supernumerary premolars.

Histopathologically, it shows hyperorthokeratinized stratified squamous epithelium with surface corrugations and the fibrocellular connective tissue.

In the absence of any other compelling factors, periodic monitoring of these teeth is preferred to surgical removal.[2] Whenever these teeth are associated with any pathological formation or when they hinder the eruption of, or give rise to a malpositioning of the permanent teeth, they should be removed as soon as possible.[9] Following these guidelines, teeth on left side were removed, whereas that on right side were not touched. Due to the very low rate of recurrence and lack of aggressiveness, enucleation is recommended as the treatment for OOC.[7]

In conclusion, a rare case of simultaneous occurrence of BMSMP and OOC associated with impacted multiple supernumerary premolars in mandible is reported. Though the percentage of pathologies associated with impacted supernumerary premolars is less; in our opinion, if feasible impacted supernumeraries should be removed to prevent future morbidity.

References

1. Saini T, Keene JJ, Whetten J. Radiographic diagnosis of supernumerary premolars: Case reviews. J Dent Child 2002;69:184-90.
2. Farahani RM, Zonuz AT. Triad of bilateral duplicated permanent teeth, persistent open apex, and tooth malformation: A case report. J Contemp Dent Pract 2007;8:94-100.
3. Hopcraft M. Multiple supernumerary teeth- case report. Aust Dent J 1998;43:17-9.
4. Hyun H, Lee S, Ahn B, Lee S, Heo M, Seo B, *et al*. Nonsyndromic multiple mandibular supernumerary premolars. J Oral Maxillofac Surg 2008;66:1366-9.
5. Rubenstein L, Lindauer S, Isaacson R, Germane N, Richmond V, Augusta G. Development of supernumerary premolars in an orthodontic population. Oral Surg Oral Med Oral Pathol 1991;71:392-5.
6. Kantor ML, Bailey S, Burkes JE. Duplication of the premolar dentition. Oral Surg Oral Med Oral Pathol 1988;66:62-4.
7. Dong Q, Pan S, Sun L, Li T. Orthokeratinized odontogenic cyst-a clinicopathologic study of 61 cases. Arch Pathol Lab Med 2010;134:271-5.
8. Bhatia R, Mehta D. Familial occurrence of non-syndromal multiple supernumerary teeth: A rare case report. J Indian Dent Assoc 2010;4:241-3.
9. Kaya GS, Yapici G, Omezli MM, Dayi E. Non-syndromic supernumerary premolars. Med Oral Patol Oral Cir Bucal 2011;16:e522-5.
10. Solares R, Romero M. Supernumerary premolars: A literature review. Pediatr Dent 2004;26:450-8.
11. Yusof WZ. Non syndrome multiple supernumerary teeth: Literature review. J Can Dent Assoc 1990;56:167-9.

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