Multiple Calcifying Hyperplastic Dental Follicles: A Case Report

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Multiple calcifying hyperplastic dental follicles (MCHDFs) are a rare condition of calcification in the follicles of multiple impacted teeth. Radiologically, they appear as multiple pericoronal radiolucencies with radiopaque foci. This report presents a case of MCHDFs in a 22-year-old man.

J Oral Maxillofac Surg 77:757-761, 2019

Multiple calcifying hyperplastic dental follicles (MCHDFs) are a rare condition involving the follicles of multiple impacted teeth.[1] Radiologically, they appear as multiple pericoronal radiolucencies with radiopaque foci. Histologically, MCHDFs are similar to central odontogenic fibromas and calcifying epithelial odontogenic tumors (CEOTs).[2,3]

To the authors' knowledge, 15 cases of MHCDFs have been reported in the English-language scientific literature to date.[1,4-13]

Report of Case

A 22-year-old man was referred to the Maxillofacial Surgery Department of the Al-Zahra University Hospital (Isfahan, Iran) in 2015. He was previously examined by a general dentist for a chief complaint of multiple edentulous areas in the oral cavity. After requesting panoramic radiography, the dentist noted several impacted teeth with mixed radiopaque radiolucent pericoronal lesions (Fig 1). Therefore, he referred the patient to the maxillofacial surgery department.

The patient's medical and family histories were unremarkable. He did not use any medication or drugs. The patient did not report pain or paresthesia in his jaws and no extraoral swelling of the jaws was present.

Intraoral inspection showed distinct firm prominent areas on the buccal surface of the maxilla and mandible. The patient was referred for cone-beam computed tomography (CBCT).

CBCT FINDINGS

According to the CBCT scans (Figs 2 and 3), the upper right central and lateral incisors, first premolar, first molar, and deciduous second molar; upper left central and lateral incisors, first premolar, and first molar; lower left central and lateral incisors, first molar, and deciduous second molar; and lower right central and lateral incisors, first molar, and deciduous second molar were erupted. The upper right canine, second premolar, and second and third molars; upper left canine, second premolar, second and third molars, and deciduous second molar; lower left canine, first and second premolars, and second and third molars; and lower right canine, first and second premolars, and second and third molars were impacted. There were well-defined mixed lesions surrounding the crowns of the impacted teeth. The lesions had caused displacement of the involved teeth. The buccal and lingual cortices were also expanded and thinned by the lesions in both jaws. Displacement of the inferior alveolar canal by the lesions in the mandible also was present. Moreover, the lesions had caused loss of continuity in the floor of the nasal fossa. According to the radiographic findings, the differential diagnosis included multiple hyperplastic follicles, multiple dentigerous cysts, and

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Conflict of Interest Disclosures: None of the authors have any relevant financial relationship(s) with a commercial interest.

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Received August 4 2018

Accepted November 21 2018

0278-2391/18/31287-4

https://doi.org/10.1016/j.joms.2018.11.021

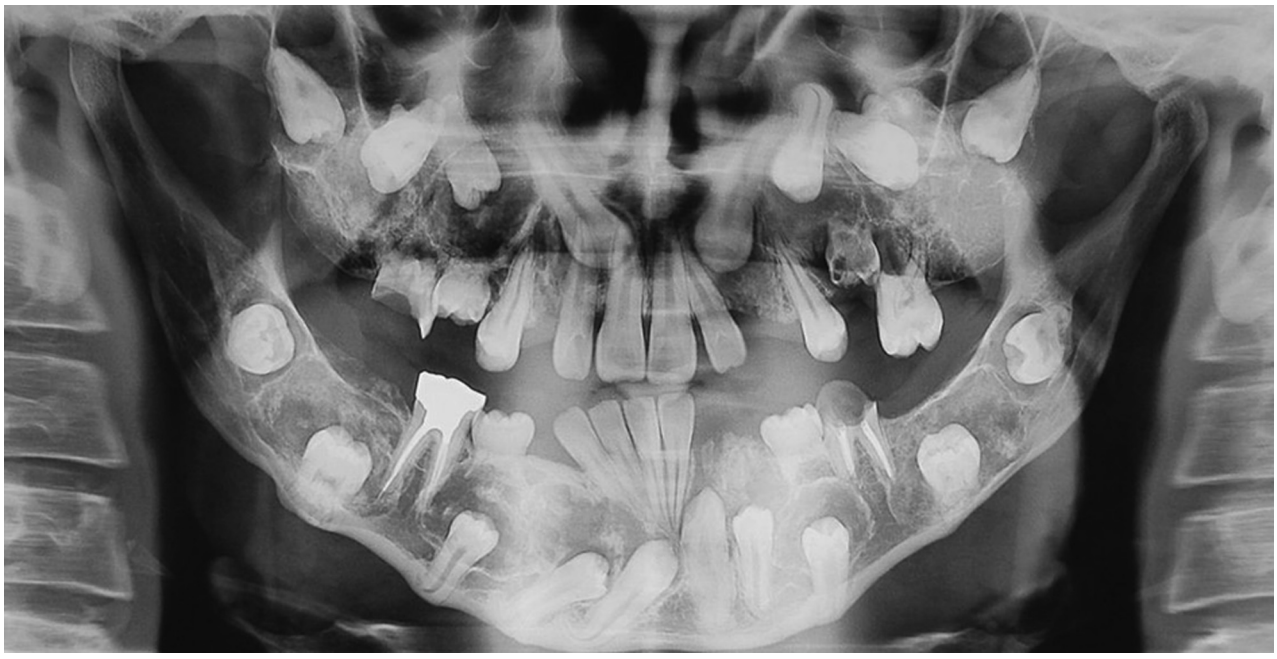

FIGURE 1. Panoramic radiograph of patient showing multiple mixed radiopaque radiolucent pericoronal lesions.

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multiple keratocystic odontogenic tumors. Also, owing to the probability of Gardner syndrome, the patient was referred to a gastroenterologist. In addition, blood chemistry test results were normal. Excisional biopsy was selected as the treatment plan.

SURGICAL PROCESS

Three surgical sessions were performed: 1 for the left side of the mandible, 1 for the right side of the mandible, and 1 for the maxilla. In each session, after preparation of the patient and under general anesthesia, a sulcular incision was performed in the region. After elevation of the flap and exposure of the underlying bone, the lesions and impacted teeth were exposed with a rotary round bur. Lesions were enucleated and curetted. The gross enucleated specimen contained multiple cream-and-brown lesions. Then, the incision area was closed with suturing.

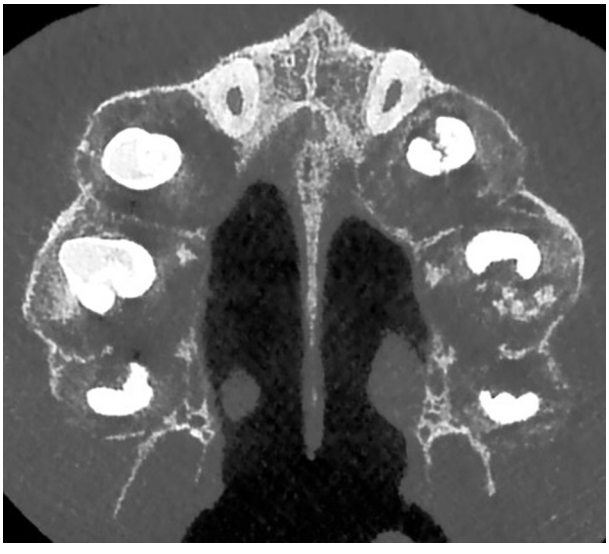

FIGURE 2. Axial cone-beam computed tomographic view of lesions in the maxilla.

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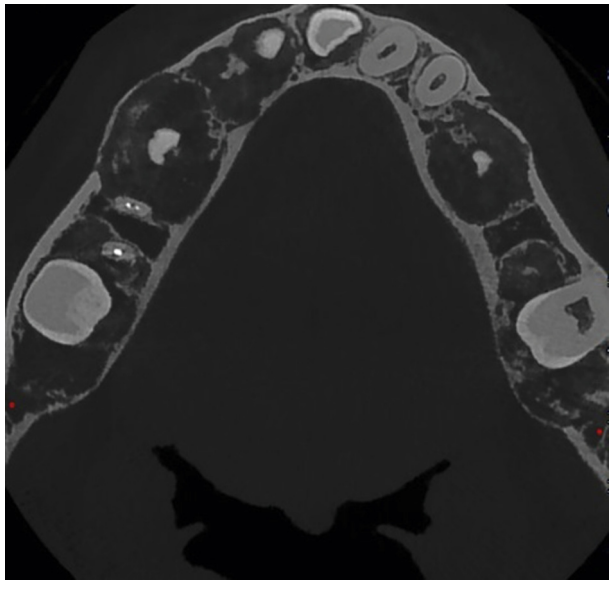

FIGURE 3. Axial cone-beam computed tomographic view of lesions in the mandible.

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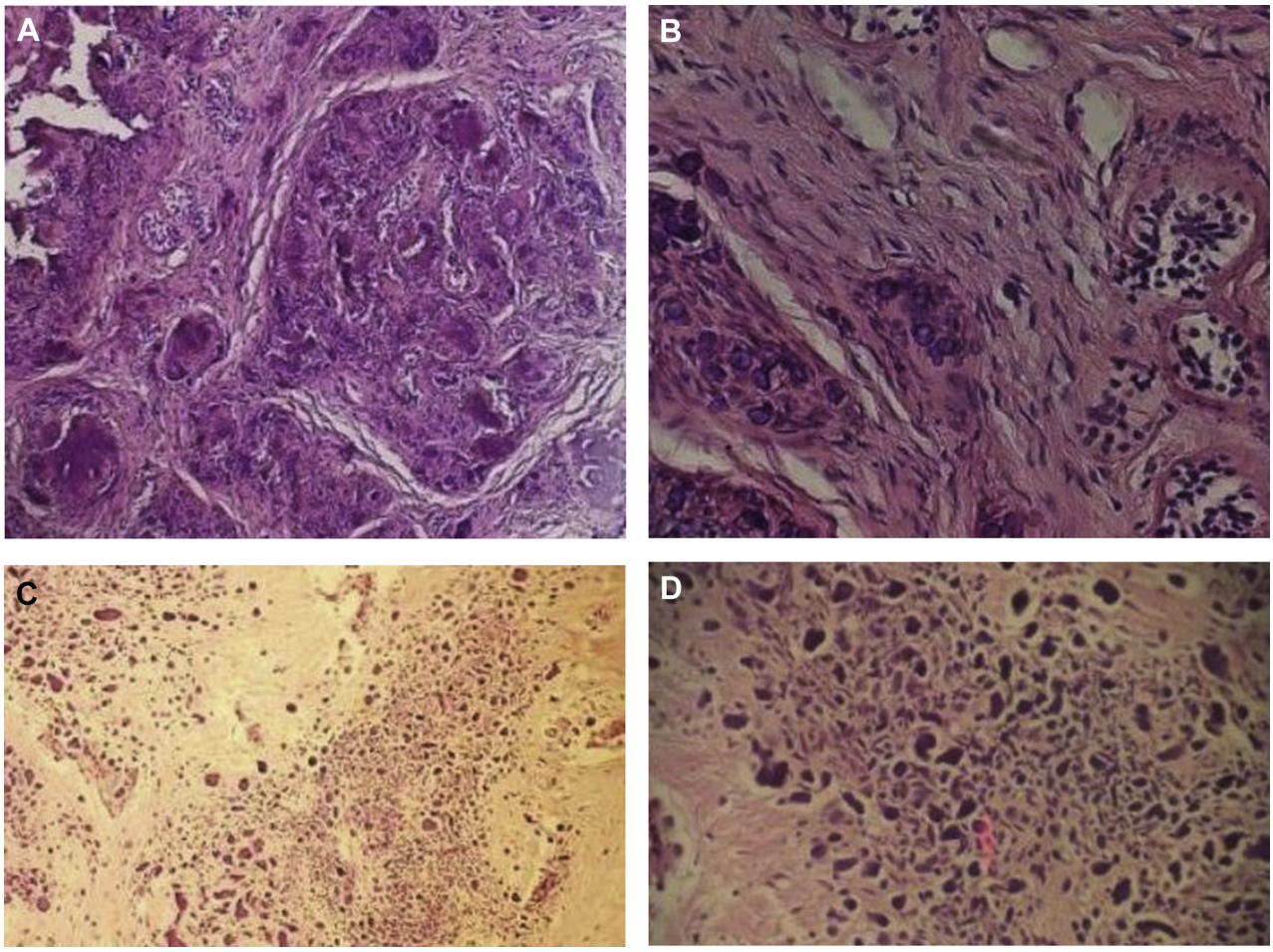

FIGURE 4. Microscopic lesion specimen. *A*, Dental follicles show cellular connective tissue with type I calcification arranged in whorled structures (hematoxylin and eosin stain; magnification, ×100). *B*, Representative section displays dense fibrous connective tissue with a whorled pattern. Islands of odontogenic epithelium with numerous clear cells are visible (magnification, ×400). *C*, Type II calcification with peripheral tufts within the fibrous connective tissue stroma (magnification, ×100). *D*, Type II calcification (magnification, ×400).

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PATHOLOGIC FINDINGS

In the microscopic specimen (Fig 4), fibrous connective tissue containing fibroblasts and collagen fibers with epithelial cords and islands were observed. Clear cells also were seen in the specimen. Type I (calcification arranged in a whorled structure) and type II (calcification with peripheral tufts within the fibrous stoma) calcifications were present, although type I calcification was predominant. Congo red staining was negative, thus ruling out CEOT from the diagnosis.

Some studies advocate picrosirius red stain for distinguishing MCHDFs from central odontogenic fibromas. In the present case, this staining test was not performed because of clinical evidence in support of MCHDFs.

Discussion

MCHDFs are a rare condition whose cause and related factors are not clearly understood.[6,12]

Histologically, the differential diagnosis involves CEOTs, central odontogenic fibromas, and fibro-osseous lesions.[2,3] Calcifications in MCHDFs originate from mesenchymal cells of dental follicles. These cells can convert to cementoblasts and osteoblasts and produce calcified material.[5] Fifteen cases of MCHDFs have been reported in the literature. Some literature reviews on the subject have included cases of multiple pericoronal central odontogenic fibromas owing to similarities to MCHDFs. However, in the present report, only cases of MCHDFs are included (Table 1). In these reported cases, most patients were male, showing a strong gender predilection. The condition is usually observed in young patients (mean age, 18 yr). Moreover, the lesions mostly involve the mandible.[8,9] However, in the present case, the maxilla and mandible were involved. Type I calcification was reported in all cases. Type I and II calcifications were found in 8 previous cases and in the present case.

Table 1. CHARACTERISTICS OF PREVIOUS CASE REPORTS OF MULTIPLE CALCIFYING HYPERPLASTIC DENTAL FOLLICLES

Number	Year	Study	Gender	Age	Impacted Teeth, N	Type of Calcification	Connective Tissue
1	1988	Sandler et al[1]	M	15	9	I	Loose to dense fibrous
2	1990	Lukinmaa et al[4]	M	24	3	I	Dense collagenous
3	1995	Gardner and Radden[5]	M	40	6	I and II	Dense fibrous
4	1998	Gomez et al[6]	M	15	17	I and II	Dense fibrous
5	2008	Roquebert et al[7]	M	10	?	I and II	Dense and loose fibrocollagenous and myxoid
6	2011	Cho et al[8]	M	11	7	I and II	Myxoid to loose fibrous
7			M	14	6	I and II	Myxoid to loose fibrous
8			M	11	4	I	Myxoid to loose fibrous
9			M	15	7	I	Dense fibrous
10			M	17	3	I and II	Dense fibrous
11	2013	Jamshidi et al[9]	M	19	2	I and II	Fibrous
12	2013	Aydin et al[10]	F	31	6	I and II	Loose to moderately dense collagenous
13	2014	O'Connell et al[11]	F	12	10	I	Fibrous and fibromyxoid
14	2015	Shirafkan et al[12]	M	13	15	I	Dense and cellular fibrous
15	2017	Desai et al[13]	M	16	12	I	Dense and cellular fibrous
16	2018	present case	M	22	19	I and II	Dense fibrous

Abbreviations: F, female; M, male.

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Considering the greater male tendency of MCDHFs, genetic and hormonal causes could be considered etiologic factors. Although the exact etiology of MCHDFs is not clearly understood, further genetic studies could clarify some etiologic aspects of the condition. Multiple impacted teeth occur in syndromes such as Gardner syndrome, Gorlin-Goltz syndrome, and cleidocranial dysplasia.[5,14,15] Gorlin-Goltz syndrome and cleidocranial dysplasia were ruled out by clinical information.

Controversy exists in the literature regarding the nature of multiple calcified pericoronal radiolucencies. Some investigators have stated that these lesions are central odontogenic fibromas, whereas others have claimed that they are calcified hyperplastic dental follicles. This difference in terminology most likely derives from the pathologist's inability to distinguish central odontogenic fibromas from hyperplastic dental follicles. However, central odontogenic fibromas are generally larger and associated with tumor-like characteristics, such as bone expansion.[7]

Because of serious complications accompanied by Gardner syndrome, consultation with a gastroenterologist was sought and he postponed more inspections to after oral and maxillofacial surgery.

Twenty-four hours after surgery, marked ecchymosis and swelling appeared. These symptoms disappeared completely after 1 month. Temporary paresthesia occurred but resolved after 3 months.

When multiple impacted teeth with enlarged follicles are seen at initial radiography, the clinician must suggest appropriate imaging modalities such as CBCT for further detailing the characteristics of the lesions. In such cases, MCHDFs must be included in the differential diagnosis.

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