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## Case report

# An unusual case of focal cemento-osseous dysplasia occupying nearly the entire maxillary sinus and arising from adjacent tissue of fused teeth

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## ABSTRACT

Cemento-osseous dysplasia (COD) of the jaw is a benign fibro-osseous lesion that is subdivided into three types, periapical, focal, and florid, based on the clinical and radiologic features. Focal COD (FCOD) usually affects the posterior tooth-bearing area or edentulous alveolar process and typically presents as an asymptomatic lesion diagnosed incidentally on dental radiography. In asymptomatic cases, once the diagnosis has been established, invasive treatment is not required. However, routine follow-up is warranted, because some patients develop multiple-site involvement, i.e., florid COD. Symptomatic patients require treatment. We describe the unusual case of a 19-year-old Japanese woman with FCOD in the left maxillary sinus who presented with repeated discomfort in the left side of the cheek. Radiographic examinations showed a mixed radiolucent-radiopaque, well-defined maxillary mass that obliterated nearly the entire left maxillary sinus. We planned to remove the lesion to restore the nasal ostium patency and natural sinus clearance mechanism, because clinical symptoms were considered to result from obliteration of the maxillary sinus by the lesion. The lesion was enucleated via the intraoral approach under general anesthesia. The microscopic examination showed a dense hypocellular sclerotic mass composed of compact bone-like cemento-osseous tissue with irregular lamellar structures. The final diagnosis was determined to be FCOD considering the combination of clinical, radiologic, and histopathological findings. The clinical outcome was satisfactory without postoperative infection or complications.

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## 1. Introduction

Cemento-osseous dysplasia (COD) is a type of fibro-osseous lesion (FOL) of the jaw, the presence of which indicates idiopathic processes in the periapical region of the tooth-bearing jaw areas characterized by replacement of normal bone by fibrous tissue and metaplastic bone.

The 2017 World Health Organization (WHO) Classification of Head and Neck Tumors [1] subdivided COD into three groups: periapical COD affecting the anterior mandible and involving one or more teeth; focal COD (FCOD) with characteristics similar to periapical COD but with a limited number of lesions in the posterior jaw quadrant; and florid COD localized primarily to the tooth-bearing or edentulous areas and often affecting patients bilaterally with symmetric involvement. The diagnosis of each type of COD usually is based on clinical and radiologic features, because the microscopic features are similar among all three types of COD and, to some extent, depend on the degree of calcification of the lesions [2,3]. These lesions are clinically asymptomatic and usually found during routine dental radiographic examinations, which show specific

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radiographic features associated with their development stages [4]. Although the pathogenesis and etiology of COD are uncertain, some reports have indicated that the pathology originates from the periodontal ligament tissue [2]. COD generally is not considered to be comprised of odontogenic lesions. However, almost all appear above the mandibular canal or below the junction of the hard palate and, thus, are confined to the alveolar process [5]. Although all types of COD occur predominantly in the mandibular or maxillary periapical lesions or a previous extraction site, it is extremely rare that the COD occupies nearly the entire maxillary sinus [6]. Moreover, FCOD related to fused teeth of the impacted third and supernumerary microdontia in a maxilla has not been reported.

We present a case of FCOD occupying nearly the entire maxillary sinus and associated with impacted fused teeth.

## 2. Case report

A 19-year-old woman was referred to our hospital with the complaint of repeated facial pain over the maxillary region with nasal discharge. The detailed history showed that about 6 years previously the patient felt slight discomfort in the left cheek. A panoramic radiography scan showed a large radio-opaque lesion in the left maxillary sinus.

No swelling of the left cheek or facial asymmetry was observed. Intraoral examinations showed no tender swelling from the left upper premolar region to the maxillary tuberosity corresponding to the buccal region. The patient's medical and family histories were not remarkable. The results of routine blood examinations were within the normal range.

The lesion was assessed preoperatively using panoramic radiography and computed tomography (CT). A panoramic radiograph showed a well-defined and mixed radiolucent-radiopaque lesion in the left maxillary sinus (Fig. 1). The CT scan showed a massive heterogeneous lesion approximately 36 mm in diameter, which involved the impacted third molar apical area and protruded into the maxillary sinus. The left maxillary sinus cavity was occupied by the mass and thickened mucosa. The CT scan also confirmed that the roots of both the impacted third molar and the supernumerary fourth molar were fused (Fig. 2). On the basis of clinical and radiographic examinations, a clinical diagnosis of "Cemento-osseous dysplasia" was made.

We excised the left maxillary tumor and the impacted fused molars under general anesthesia. A full-thickness mucoperiosteal flap was formed from the medial side of the upper left second incisor to the distal side of the upper second molar. The lateral wall of the maxillary sinus was opened with an osteotome and a mallet to excise the tumor. The tumor was white and roughly round in shape with a diameter of about 30 mm, had a granular texture and was attached to impacted fused molars. The tumor was excised with a dental handpiece and a straight bur. The impacted fused molars were extracted. The soft tissue lining of sinus appeared healthy and hence was not removed. Macroscopic examination of the resected materials showed gritty bone tissue fragments with slight hemorrhagic changes (Fig. 3A). Microscopically, formation of a dense hypocellular sclerotic mass with expansive growth causing thinning of adjacent cortical bone was seen (Fig. 3B). The lesion was comprised of compact bone-like cemento-osseous tissue with irregular lamellar structures (Fig. 3C). The space containing slight fibrous component between compact bone-like cemento-osseous mass and thinning adjacent cortical bone can be identified. There were slight fibro-osseous components which might be in clinically radiolucent area in fragmented resected materials (Fig. 3D). Regarding the impacted fused tooth, the macroscopic view showed that the third molar was truly fused to the supernumerary microdontia and the two shared dentin (Fig. 4A, B). Root resorption of the fused

tooth was observed. On the cut surface of the fused tooth, a cavity of dental pulp was shared by both teeth (Fig. 4C).

Based on the clinical, radiologic, and histologic findings, a diagnosis of focal cemento-osseous dysplasia was established. Postoperative panoramic radiography showed complete removal of the lesion (Fig. 5). The postoperative follow-up period was uneventful, and the patient was discharged from the hospital 5 days postoperatively. After discharge, no relapses or postoperative complications occurred during the 2-year follow-up period.

## 3. Discussion

The fibro-osseous lesions (FOLs), conditions that affect the jaw and craniofacial bone, are characterized by replacement of normal bone by a benign cellular connective tissue matrix [5]. The FOLs in the maxillofacial region include COD, fibrous dysplasia (FD), ossifying fibroma (OF), and their subtypes [7]. The group comprises a wide range of conditions, the correct diagnosis and classification of which can be challenging.

COD is a non-neoplastic lesion included in the "Fibro-osseous lesion and osteochondromatous lesions" listing in the WHO Classification of Head and Neck Tumors with FD and OF [1]. The WHO classification also described three clinical presentations of COD, i.e., periapical COD, FCOD, and florid OD.

In 1994, Summerlin and Tomich suggested the term focal osseous dysplasia (FOD) (currently a synonym for FCOD) primarily based on localization of cemental dysplasia [8]. FOD is the most common FOL of the jaws and is seen predominantly in women, with a peak incidence during the fourth and fifth decades of life; FOL affects the posterior mandibular tooth-bearing area, with most lesions smaller than 20 mm [6,9]. The lesions also are confined to edentulous spaces at the site of previous extractions [3]. The incidence rate of FOD in the maxilla is limited to 20% of cases [6]. The current data agreed with information in the literature regarding patient sex but not age and location. In the current case, the location and size differed from other reports. Thus, the similarities are few [10].

Patients with FOD are generally asymptomatic and do not require treatment unless infection or symptoms occur. In contrast, a report of 54 Japanese patients with OD showed that 32 (59%) patients presented with at least one sign and symptom of inflammation [11]. It is difficult to control infection with OD by administration of antimicrobial agent, because the lesions are in an avascular area. Our patient had pain in the cheek that could have been attributed to chronic sinusitis owing to obstruction of the natural ostia by the massive lesion. Therefore, we performed surgery to remove the lesion that obstructed the natural ostia.

A wide range of radiographic appearances of FOD with three developmental stages has been reported in previous studies [12]. Because of its progressive mineralization, the radiographic appearance depends on the developmental stage. The early-stage lesions present radiographically with a periapical well-defined radiolucent defect. In this stage, the lesions might be diagnosed erroneously as an endodontic lesion. The intermediate-stage lesions have mixed or pure radiodensity with a radiolucent rim around the radiopacity. In contrast, some cases at this stage had an ill-defined mixed radiodensity without a lucent rim. The late-stage lesions display mainly a sclerotic radio-opacity with an ill-defined border. The current case was considered late stage because the findings on imaging showed predominant radio-opacity in the left maxillary sinus, and the pathological findings indicated that the lesion consisted predominantly of dense hypocellular sclerotic cemento-osseous tissue. Because COD are a group of disorders known to originate in the periodontal ligament tissues [11], the lesion was considered

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