

Cemento-Osseous Dysplasia-Like Lesion and Complex Odontoma Associated with an Impacted Third Molar

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Abstract We present a case of a 36-year-old female with a benign fibro-osseous lesion consistent with cemento-osseous dysplasia (COD) that was located coronally to the crown of an impacted mandibular third molar tooth and was associated with a complex odontoma. The pathogenesis of this unusual association is discussed and the odontogenic origin of COD is suggested.

Keywords Benign fibro-osseous lesions ·
Cemento-osseous dysplasia · Complex odontoma ·
Impacted tooth

Introduction

Benign fibro-osseous lesions of the craniofacial complex comprise a diverse group of processes characterized by replacement of normal bone by hypercellular fibrovascular connective tissue that produces mineralized osteoid or cementum-like material [1, 2].

Cemento-osseous dysplasia (COD) encompasses the most common fibro-osseous lesion and is subclassified into focal, periapical, and florid subtypes [2, 3]. They generally

occur in the tooth-bearing areas of the jaws and may be localized or multiple. The localized periapical and focal COD subtypes occur predominantly in the anterior and posterior mandible, respectively, of middle-aged females and are small, asymptomatic lesions. Florid COD is characterized by lesions in multiple quadrants of the maxilla and mandible. Radiographically, early, intermediate, and late stages are recognized by progression from radiolucent, to mixed radiolucent-radiopaque, to radiopaque lesions with well-defined borders, respectively [3].

Association of benign fibro-osseous lesions of the jaws with simple bone cysts [4] and aneurysmal bone cysts [5, 6] has been previously reported. Herein, we present a case of a benign fibro-osseous lesion consistent with COD that was located coronally to the crown of an impacted mandibular third molar, and which was also associated with a complex odontoma.

Case Details

A panoramic radiograph in a 36-year-old female complaining of pain in the region of the maxillary right second premolar and first molar teeth, incidentally revealed a deeply impacted mandibular left third molar. The proximal molar tubercle of the tooth had an indistinct outline and the root was irregularly-shaped, short, and projecting into the inferior alveolar canal. The crown was partly surrounded by a mixed radiopaque-radiolucent lesion with ill-defined borders that was more radiopaque peripherally and more radiolucent centrally (Fig. 1). These findings were suggestive of a complex odontoma. The patient's medical history was non-contributory and she was not taking any medications.

On clinical examination, the mandible appeared normal, without expansion of the cortical plates, and was covered

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Fig. 1 Panoramic radiograph shows the crown of the impacted mandibular left third molar surrounded by a mixed radiopaque-radiolucent lesion with ill-defined borders

by normal mucosa. With a preliminary working diagnosis of “impacted third molar and complex odontoma” the lesion was excised, along with the second and third molars. Due to the lack of encapsulation, the lesion was removed by curettage, and an osteosynthesis plate was fixed in place. Notably, no recurrence has been recorded after two years.

Grossly, the biopsy consisted of multiple gray-white, soft to hard tissue fragments, measuring in aggregate 2.0 cm in greatest dimension. The specimen was fixed in 10% buffered formalin and decalcified in Surgipath decalcifier II® (Leica Microsystems). The paraffin embedded tissue was cut in 5 μ m-thick sections and stained with hematoxylin and eosin.

Microscopic examination revealed two distinct yet intermingling lesions (Fig. 2). The first was a typical complex odontoma characterized by haphazardly arranged dentin enveloping odontogenic epithelium and pulpal tissue (Fig. 3). The second lesion featured intersecting bundles of cellular, fibrovascular, connective tissue with numerous irregularly shaped calcifications akin to bone or cementum (Fig. 4a). Adjacent areas showed thick, curvilinear, and hypo-cellular bone trabeculae with basophilic outlines and rare osteoblastic rimming in a background of vascular, but sparsely cellular connective tissue (Fig. 4b). Diffuse interstitial hemorrhage was present. Although unencapsulated, the lesion was surrounded by mature bone. A final diagnosis was made of benign fibro-osseous lesion, consistent with COD, in association with complex odontoma.

Discussion

The co-existence of a benign fibro-osseous-like lesion, consistent with COD, and an odontoma around an impacted

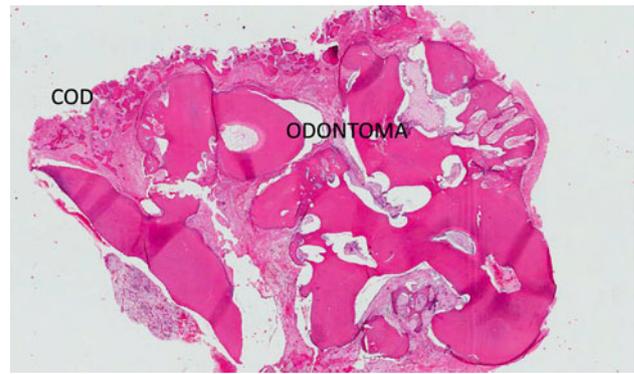


Fig. 2 Areas of odontoma (OD) and COD-like (COD) tissue (hematoxylin and eosin stain, original magnification $\times 10$)

tooth in the present case may be explained either by (1) the presence of COD-like features in an odontoma, (2) coincidental association of two distinct lesions, or (3) a common developmental origin for both lesions.

Odontomas, the most common odontogenic tumors, are divided into compound and complex types, respectively. The latter subtype, as seen in this case, has a radiopaque appearance on imaging and is characterized by a haphazard arrangement of dental tissues. Approximately half of odontomas arising in the mandibular molar area are associated with impacted teeth [7]. Small amounts of cementum material are rarely found in odontomas and surrounding bone trabeculae may appear in the surgical specimen. However, areas with typical features of COD, as seen in this case, have not been previously described.

CODs, which are more common in the apical aspect of teeth, do not show enamel or dentin [1]. The only established association of COD with another lesion is with simple bone cysts [4]. Interestingly, Lida et al. [8] reported an unusual coexistence of COD with odontoma in a 37-year-old Japanese female with multiple fibro-osseous lesions in the same jaw quadrant. This patient had fibro-osseous lesions that “enveloped” an impacted maxillary canine, and involved the apices of a partly erupted second molar and impacted third molar, respectively. This latter tooth was in close proximity to an odontoma within a dentigerous cyst. Due to the multifocal presentation of the fibro-osseous lesions on the same maxillary side, the diagnosis of multiple focal COD was made. However, the lesion surrounding the upper canine had caused bone expansion, thus behaving more like a neoplasm. In view of this, coincidental association of COD with odontoma would be very unusual, especially taking into consideration the coronal location of the COD.

Odontomas are thought to be hamartomas arising from differentiated odontogenic epithelial and ectomesenchymal cells, as a result of local trauma, infection, or genetic factors [7, 9]. In contrast, CODs are thought to develop

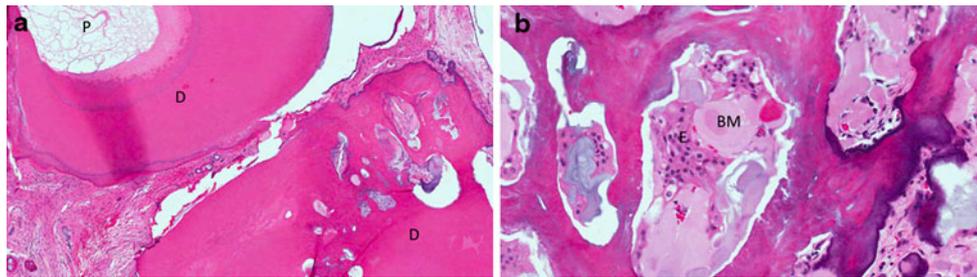


Fig. 3 Odontoma showing (a) masses of haphazardly arranged dentine (D) enclosing pulp-like tissue (P) and (b) odontogenic epithelium (E) in abortive enamel matrix (BM) (hematoxylin and eosin stain, original magnifications $\times 50$, $\times 200$)

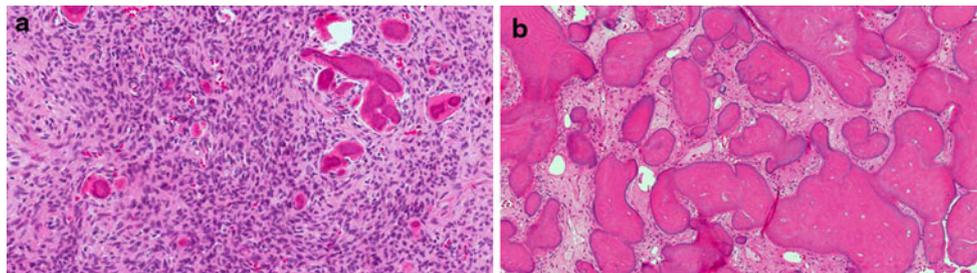


Fig. 4 COD-like lesion showing (a) cellular fibrovascular connective tissue with cementum-like material and (b) anastomosing trabeculae of bone in sparsely cellular connective tissue (hematoxylin and eosin stain, original magnifications $\times 100$)

from the periodontal ligament (periapical COD) or the medullary jaw bone (focal COD) [2]. In this case, origin from the periodontal tissues of the second molar that was in close proximity to the lesion is not likely because of the intact periodontal space and the association of the second molar with the radiopaque (odontoma) and not with the radiolucent (COD-like) part of the lesion. Moreover, if COD-like lesions were a localized reaction, due to trauma or infection from the bone surrounding the impacted tooth and/or odontoma, one would expect more CODs to arise in a coronal location and in association with odontomas, since both tooth impaction and odontomas are common.

The common link between both lesions, in this case, may be the dental follicle of the impacted third molar. In fact, hyperplasia of the follicular tissue in regional odontodysplasia may create a COD-like lesion, with interlacing bundles of cellular connective tissue, basophilic enamel-like calcification (enameloid conglomerates), and islands of odontogenic epithelium [10]. However, although our findings of an indistinct (“fuzzy”) outline of the proximal coronal tubercle and the short, misshaped root could represent a localized form of regional odontodysplasia, the overall clinical, radiographic, gross, and microscopic features are not considered diagnostic of this entity. Origin from the dental follicle would indicate that COD is odontogenic, a presumption that is also supported by more

frequent occurrence of COD in the tooth-bearing areas of the jaws [1, 2].

Although the association of COD-like lesions and odontoma may seem solely of academic interest, it suggests an odontogenic origin of COD. In order for this to be substantiated, more reports of COD-like lesions arising in association with odontogenic tumors are needed.

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