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Benign fibro-osseous lesion and odontoma of the mandible: A report of a rare, hybrid lesion and review of literature

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ABSTRACT

This report documents a case of a 12-year-old male with a combined benign fibro-osseous lesion (BFOL) and odontoma. Radiographic imaging revealed an extensive, well-defined, multilocular, radiolucent lesion with small radiopaque entities in the left mandible. Microscopic examination revealed a central BFOL with associated odontomas. To the best of the authors' knowledge, this is the third report of a combined BFOL and odontoma and an extensive review of the English language literature on combined BFOL and odontoma cases is included. This case is important because it demonstrates the importance of diagnosing bone lesions with an accompanying radiograph. The initial biopsy was signed out as a BFOL with a comment that there was tooth-like material as well and a possibility that it represented an odontoma. The patient was operated on in our teaching hospital and the lesion completely excised with a diagnosis of dentigerous cyst. The hard tissue was grossed only as it appeared tooth-like. Seeing the final diagnosis, we requested decalcification of the hard tissue. Based on clinical, radiographic and histologic findings, the final diagnosis of BFOL and odontoma was made. Another reason for reporting this case is that the paucity of published cases of these combined lesions makes treatment decisions a challenge. Further future reporting of BFOL arising in association with odontomas with long-term follow-up information is paramount to better understand these entities vis-à-vis their nature and behavior.

1. Introduction

Benign fibro-osseous lesions (BFOL) of the jaws are a diverse group of lesions characterized by the replacement of normal bone with fibrous tissue containing variable amounts of mineralized product [1]. Some examples include fibrous dysplasia, cemento-osseous dysplasia (COD), and cemento-ossifying fibroma (COF). These entities exhibit similar histopathologic features, but differ in etiology, biologic behavior, treatment, and prognosis.

Odontomas are a common developmental anomaly derived from odontogenic epithelial and mesenchymal tissues [1]. They consist of enamel and dentin with variable amounts of pulp and cementum. Two variants of odontomas exist: compound odontomas and complex odontomas. Compound odontomas are composed of multiple, small tooth-like structures whereas complex odontomas consist of a conglomerate mass of enamel and dentin that bear no anatomic resemblance to a tooth [1]. Odontomas may occur in any tooth-bearing area of the jaws, with the compound type mostly seen in the anterior maxilla and complex odontomas frequenting the posterior mandible [1].

Herein, we present a case of a BFOL and associated odontomas that were preventing the eruption of the patient's left mandibular molars. This is the third report of a hybrid BFOL and odontoma.

2. Case report

A 12-year-old male presented to the University of Florida Oral Surgery Clinic with the chief complaint of missing molars on the left mandible. The patient had not seen a dentist for routine care since he was 4-years-old. His medical history was noncontributory and he was

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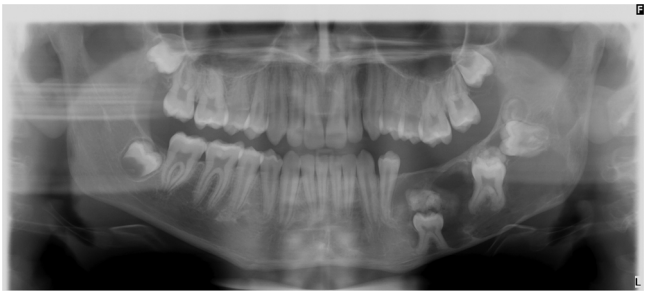

Fig. 1. Pantomograph depicting radiopaque and radiolucent areas in the left mandibular body lesion. *Color online only*.

not taking any medications. He denied a history of pain, swelling, paresthesia, facial asymmetry, or drainage. Extraoral examination was within normal limits. Intraoral examination revealed palpable buccal expansion on the left mandibular vestibule and edentulism in the area of the left mandibular posterior teeth. The left maxillary posterior teeth were supraerupted.

Evaluation of the pantomograph revealed a large, multilocular radiolucent lesion in the left mandible extending from the distal of the first premolar posteriorly to the mid-ramus of the mandible (Fig. 1). Focal radiopacities were seen within the lesion. Based on the clinical and radiographic findings, the patient was recommended to have cone beam computed tomography (CBCT) imaging and an incisional biopsy of the lesion.

The CBCT radiographic findings showed a well-defined, partly corticated, multilocular, expansile, and predominantly radiolucent entity in the left mandible extending from the distal of the first premolar to the distal of the second molar anterio-posteriorly, and from the alveolar crest to the inferior border of the mandible vertically (Fig. 2A). The first and second permanent molars and retained deciduous second molar in the left mandible were involved and appeared displaced (Fig. 2B). Small radiopaque foci with tooth-like density and morphology were present above the first permanent molar and retained

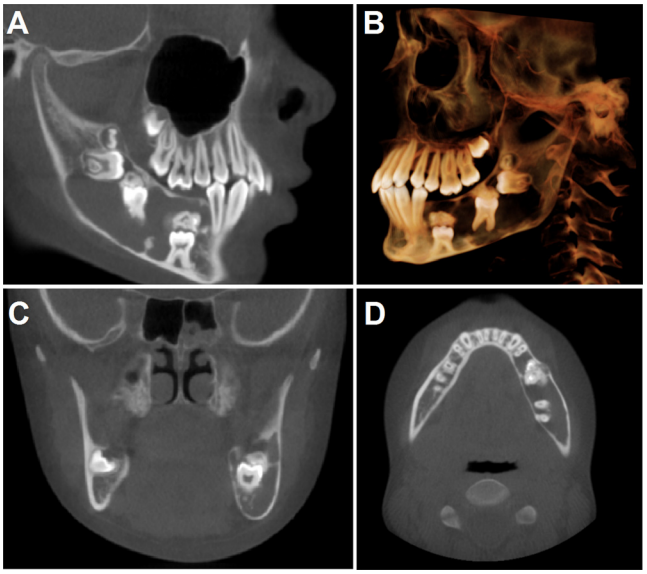

Fig. 2. Cone beam computed tomography (CBCT) imaging. (A) Sagittal view depicting a predominantly radiolucent mixed-density lesion in the mandible with multiple radiopaque entities of tooth-like morphology and density. (B) 3D reconstruction showing retained and displaced deciduous second molar and displaced permanent first and second molars. (C) Coronal view depicting an impacted tooth with a radiopaque entity of tooth-like density and morphology superior to it. (D) Axial view showing mild bucco-lingual enlargement and thinned borders. *Color online only*.

deciduous second molar in the left mandible (Fig. 2C). There was thinning of the buccal and lingual cortical plates and they appeared scalloped in some areas, perhaps depicting the attempt to loculate (Fig. 2D). The mandibular canal was not completely traceable, although the visualized portion appeared to be displaced inferiorly. The radiographic diagnosis was ameloblastic fibro-odontoma.

The lesion was aspirated, yielding a yellow, straw-colored fluid that became bloody. A crestal incision was made over the posterior left mandibular ridge and extended to the sulcus of the left mandibular first premolar. A full-thickness mucoperiosteal flap was reflected with a periosteal elevator #9. A round bur and handpiece were used to create a 2.0 × 1.0 cm window through the bone. Upon exploration, a hollow cavity with no cystic lining was noted. The walls of the defect were curetted with the spoon-end of the Woodson elevator and the incisional biopsy was submitted for microscopic examination. The bony cavity and flap were copiously irrigated and the flap was reapproximated and sutured with 3-0 chromic gut. The patient tolerated the procedure well with no complications.

Microscopic examination of the incisional biopsy revealed multiple sections of a specimen composed of cellular fibrous connective tissue containing calcospheritic masses (Fig. 3A). The calcospheritic masses were predominantly composed of cementum-like material (Fig. 3B). However, some areas of lamellar bone were seen as well. The stroma was highly cellular with numerous fibroblasts and osteoblasts. The stromal cells had hyperchromatic nuclei. Also seen were some larger calcifications that were more lamellar and composed of predominantly normal bone (Fig. 3C). A diagnosis of BFOL was rendered, but a comment was added to the report stating that in some areas the calcifications resembled tooth-like structures (Fig. 3D), creating the possibility of a combination ameloblastic fibro-odontoma/BFOL. Complete excision and submission of the entire lesion for microscopic examination was recommended.

The patient returned for an excisional biopsy 2 months after the incisional biopsy. A 15 blade was used to make a mid-crestal incision over the left mandibular body with a distobuccal release. A full thickness mucoperiosteal flap was elevated. The lesion was unroofed with an egg bur. The impacted deciduous second molar, permanent second molar, and permanent first molar in the left mandible were identified. The lesion showed a hollow cavity with no identifiable cystic lining. The impacted teeth were troughed with a 702 bur. The impacted teeth and the calcifications above the deciduous second molar and first permanent molar in the left mandible were meticulously extracted, taking care not to stress the thin mandibular bone. The teeth and associated follicles were submitted for pathologic examination. Bone within the cavity was removed and also sent for pathology review. The surgical site was curetted and copiously irrigated. The inferior alveolar nerve was found to be intact at completion of the case. Vivgena and DBX bone graft was placed into the surgical site (Fig. 4). The graft was then covered with a large collagen membrane. The incisions were primarily closed with 4-0 vicryl.

The soft tissue and hard tissue components of the specimen were separated. Microscopic examination of the soft tissue specimen revealed delicate to dense bundles of collagenized fibrous connective tissue with a myxoid background. Also present were islands and strands of odontogenic epithelium with peripheral stromal hyalinization. Rounded fragments of primitive myxoid tissue with plump, stellate fibroblasts and a rim of columnar odontoblasts were also seen. The histology of the soft tissue specimen was suggestive of the left mandibular third molar tooth bud. The hard tissue was separated into four sections. The first section showed normal viable bone and marrow. The second section showed hypercellular fibrous tissue with calcospheritic masses composed of cementum-like material similar to those seen in the incisional biopsy (Fig. 5A). Trabeculae of woven bone with plump osteocytes and osteoblastic rimming was also present (Fig. 5B). The histology was considered to be compatible with BFOL. The third section showed aggregates of basophilic enamel matrix (Fig. 5C). In one focus, ring-like

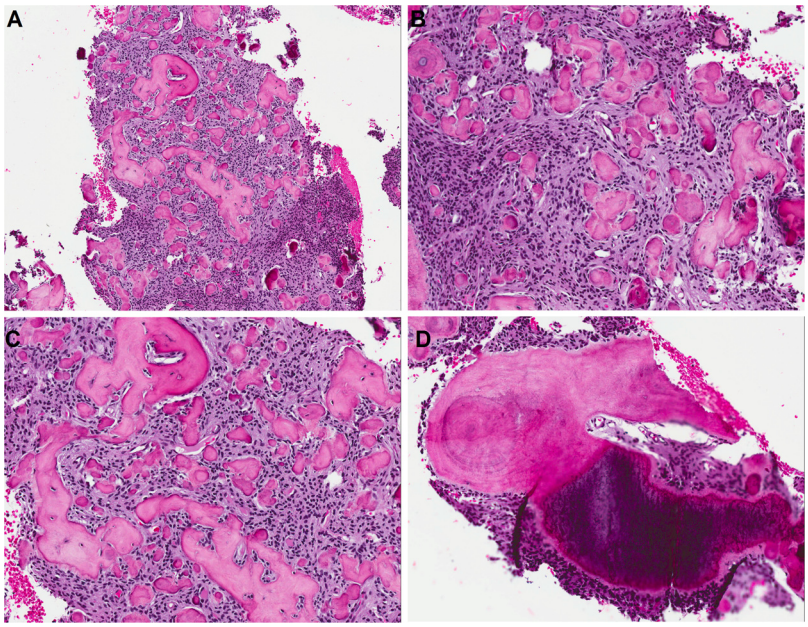

Fig. 3. Histologic representation of the incisional biopsy. (A) Cellular fibrous connective tissue with stromal cells that have hyperchromatic nuclei and calcospheritic masses (10x). (B) High-power showing cementum-like calcospheritic masses (20x). (C) Larger calcifications that were more lamellar in appearance (20x). (D) An area that resembles a tooth-like structure (20x). *Color online only*.

Fig. 4. Postoperative pantomograph demonstrating Vivgena and DBX bone graft in the left mandible. *Color online only*.

calcifications were seen (Fig. 5D). In addition, a haphazard arrangement of tubular dentin, pale eosinophilic odontogenic epithelium, and basophilic enamel matrix (Fig. 5E) with ring-like calcifications (Fig. 5F) were present. This histology was compatible with odontoma. The fourth section demonstrated two molar teeth composed primarily of dentin and myxoid, primitive dental papilla. Towards the periphery and the coronal end, enamel matrix was seen. A final diagnosis of benign fibro-osseous lesion with associated odontoma and multiple molar teeth was made.

3. Discussion

The occurrence of lesions with different histologic patterns presenting as a single lesion is extremely rare. To the best of our knowledge, this article describes the third case of a combined BFOL and odontoma within the worldwide English literature. The pathogenesis of hybrid BFOL and odontomas is currently unknown. Prodromidis et al. [2] speculates that the **co**-existence of BFOL and odontomas around impacted teeth may be explained by the presence of BFOL-like features in an odontoma, coincidental association of two distinct lesions, or a common developmental origin for both lesions. The other two cases in the literature diagnosed their lesions as cemento-osseous dyplasia (COD) associated with an odontoma [2,3]. In our case, it is difficult to specify what kind of BFOL our lesion is because it is multilocular, which is a very unusual radiographic presentation for BFOLs. Furthermore, it is well known that cemento-ossifying fibroma (COF) and COD share similar histopathologic features. According to Su et al. [4], some of these overlapping features include woven bony trabeculae intermingled among a fibroblastic stroma displaying a homogeneous or moderate cellularity. Additionally, cementum-like material of cellular and acellular spheres is equally seen in COD and COF [4]. However, distinction between these two entities is essential, as both lesions present with different clinical presentations and require different treatments.

COD is the most common fibro-osseous lesion with minimal to no neoplastic potential [1]. In contrast, COF is a true neoplasm with significant growth potential and is the most common benign fibro-osseous neoplasm of jaws [1,4]. COF can occur over a broad age range, but COD tends to occur in middle-aged to older adults [1]. In both COD and COF, there is a female predilection and the mandible is affected more often than the maxilla [1].

COD is asymptomatic and is usually detected incidentally by radiographic examination. Similar to COF, COD are rarely associated with impacted teeth. Our search of the literature revealed that the two cases of combined COD and odontoma are the only cases of COD associated with impacted teeth [2,3]. Usually COD occurs around tooth apices or in extraction sites and present either as a solitary lesion or as multiple foci [1]. Most lesions are smaller than 1.5 cm in diameter [1]. In contrast, some cases of COF may be associated with significant increase in size resulting in cosmetic and functional deformities [4]. Large lesions, especially ones that result in considerable bone destruction, may require surgical resection and bone grafting [1].

Radiographically COD varies from completely radiolucent to densely radiopaque with a thin peripheral radiolucent rim [1]. The borders tend to be well-defined to slightly irregular [1]. When biopsied, multiple fragments of small, red to tan, gritty, “gravel-like,” and friable tissue are encountered [4]. COFs, in contrast, tend to separate cleanly from bone and are removed in one or several large masses [1]. The circumscribed nature of COF allows enucleation of the tumor with relative ease [1]. The surgeons in our case did not specify if the lesion came out in multiple, gritty pieces or was enucleated in one piece. However, this is important information that should be include on biopsy submissions.

Because COD is non-neoplastic, complete surgical removal is unnecessary after a diagnosis is made [1]. The blood supply in an area of COD is usually compromised, so when elective procedures are performed, inoculation of bacteria can lead to secondary infection and suppurative osteomyelitis [5]. To avoid this complication, elective surgical procedures, such as biopsy or tooth extraction, are not

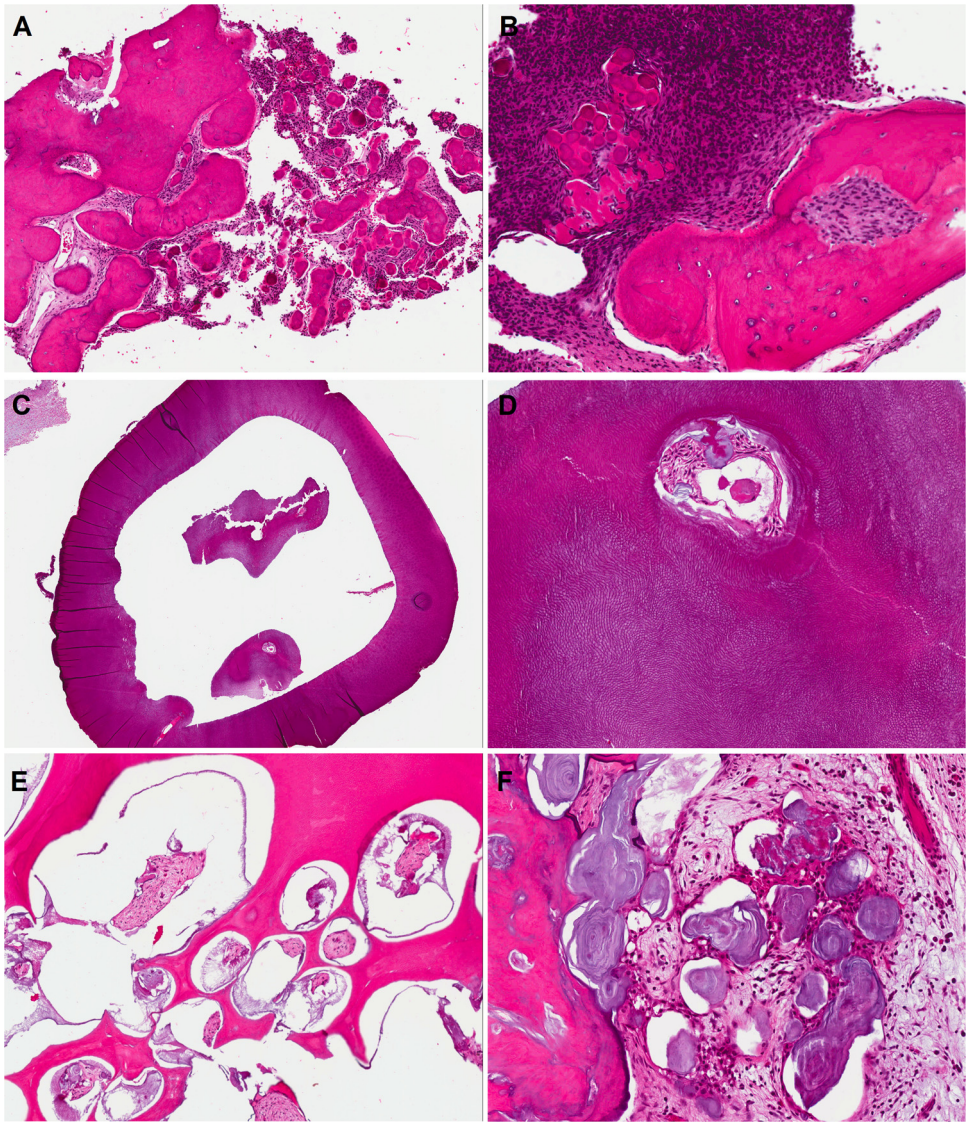

Fig. 5. Histologic representation of the excisional biopsy. (A) Hypercellular fibrous tissue with calcospheritic masses composed of cementum-like material and bone (10x). (B) Trabeculae of woven bone with plump osteocytes and osteoblastic rimming (20x). (C) Well-developed fish-scale-like enamel matrix (2x). (D) Ring-like calcification in one focus (20x). (E) Haphazard arrangement of tubular dentin, pale eosinophilic odontogenic epithelium, and basophilic enamel matrix (10x). (F) Ring-like calcifications (20x). *Color online only*.

recommended in an area of COD [1].

Though distinguishing COD from COF based on histopathologic parameters alone may be difficult, there are subtle differences that may aid with diagnosis. According to Su et al. [4], COD displays anastomosing, thick, curvilinear, relatively acellular bony trabeculae and occasional basophilic outlines forming shapes that resemble “ginger roots.” The bony trabeculae in COF, in contrast, are thin, single, and separate with frequent osteoblastic rimming [4]. The cementum-like masses in COD are located randomly throughout the specimen and are artifactually separated from the connective tissue [4]. The cemental bodies in COF are ovoid with characteristic “brush borders” that are intimately associated with the cellular components in the stroma [4]. Furthermore, storiform cellularity is more frequent, prominent, and extensive in COF [4]. However, free hemorrhage is more frequently seen and present throughout COD than COF [4]. These last two histological features are especially true for our case, as the stroma was highly cellular and lacked free hemorrhage. Based on our patient’s age as well as the radiographic presentation and histologic features of the lesion, a diagnosis of COF was favored over COD. As a result, the lesion was excised and a bone graft was placed.

Although a different BFOL diagnosis was favored in our case compared to the other two reported cases of combined BFOL and odontomas, it is still interesting to note the similarities. All three cases occurred around impacted teeth and on the left side. However, two cases occurred in the mandible and one occurred in the maxilla. The patients are all young, with an age range of 12–37 years (mean is 28-years-old). Treatment consisted of curettage or excision. A summary of all known combined BFOL and odontoma cases is presented in Table 1. None of the cases had a syndromic association. This is important to note, as individuals with Familial Adenomatous Polyposis (FAP), an autosomal-dominant colorectal cancer syndrome, may present with dental abnormalities that mimic florid COD [6].

In conclusion, bone lesions should always be diagnosed after examination of an accompanying radiograph. Combined BFOL and odontomas are extremely rare entities, but it is important to recognize them. Their unique radiographic and histologic presentations are truly worth noting as distinct lesions with unknown clinical potential. More reports of BFOL arising in association with odontomas with long-term follow-up information are needed to better understand these entities vis-à-vis their nature, clinical behavior, and possible pathogenesis.

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Table 1
Summary of combined benign fibro-osseous lesions (BFOL) and odontomas from the literature, including our current case.

Reference	Age	Gender	Location	Radiographic Presentation	Histologic Diagnosis	Treatment
Prodromidis et al. [2]	36	Female	Left mandible	Mixed radiopaque-radiolucent lesion with ill-defined borders surrounding the impacted 3^{rd} molar. The lesion was more radiopaque peripherally and more radiolucent centrally.	COD and complex odontoma	Curettage and osteosynthesis plate
Iida et al. [3]	37	Female	Left maxilla	Radiolucent lesion around the impacted 3^{rd} molar with an associated radiopaque mass.	COD and odontoma	Excision of the lesions and reduction of the surrounding alveolar bone
Current case	12	Male	Left mandible	Well-defined, partly corticated, multilocular, radiolucency surrounding the displaced and impacted deciduous 2^{nd} molar and permanent 1^{st}-3^{rd} molars. Small radiopaque entities with tooth-like density and morphology present above deciduous 2^{nd} molar and permanent 1^{st} molar. Lesion is expansile and there is thinning of the buccal and lingual cortical plates.	COF and compound and complex odontomas	Curettage and Vivgena and DBX bone graft

COD, cemento-osseous dysplasia.
COF, cemento-ossifying fibroma.

Conflict of interest

There are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

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