Cone Beam Computed Tomography (CBCT) Features of a Rare Fibro-Osseous Lesion: A Case Report

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Abstract

Cone beam computed tomography is a useful technique for imaging the craniofacial lesions. It produces more realistic images that facilitate interpretation. Juvenile ossifying fibroma (JOF) is a rare and benign fibro-osseous neoplasm that arises within the craniofacial bones, especially in the maxilla. Mandibular lesions can be seen in 10% of the cases.In both jaws, it has a predilection for the premolar and molar regions (it is mostly seen in premolar and molar regions). Radiographically, it can be present as a radiolucent, mixed or radiopaque lesion. Radiodensity varies from purely radiolucent masses to mixed densities with prominent radiopacity as the lesion matures.

This case report highlights a JOF with large foci of odontome-like radiopacities in a 6-year-old boy's mandibular anterior region. The location of the lesion in the anterior mandible and comparatively rapid formation of large odontome-like radiopaque foci at this early agehas made it a rare entity.

Key Words: CBCT, juvenile ossifying fibroma, mandible.

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Introduction

Cemento-ossifying fibroma (COF), a subdivision of fibro-osseous lesions, is a rare benign non-odontogenic tumor of the jaws (1). Confusion caused by the criteria proposed for its diagnosis, has provoked great controversy (2). Juvenile ossifying fibroma is a very aggressive form of COF which is also called aggressive or active ossifying fibroma (3). It arises within the craniofacial bonesespecially in the maxilla. Mandibular lesions are seen in 10% of the cases (4). The tumor has a predilection for (is mostly seen in) the premolar and molar regions (5). It appears at an early age and in 79% of the patients is diagnosed before the age of 15 (4,6). JOF affects both males and females equally without any significant gender predilection (7).

Radiographically, juvenile COF resembles conventional COF and classified as a clinicopathological variant of the ossifying fibroma (8).

This lesion has predominating soft tissue consistency with variable amounts of internal calcifications and/or linear or irregular focal bone (4). The amount of radiodensity varies from purely radiolucent to mixed density with prominent radiopacities. As the lesion matures, it may reveal progressive calcifications of the matrix. It usually shows a low density mass due to cystic changes on computed tomography (CT) scans (8).

Although JOF is an uncommon clinical entity, it is an aggressive lesion mimicking malignancy such as osteosarcoma. So, it is important to accurately recognize

JOF for making the diagnosis and managing the disease (9,10). The essential characteristics of this clinical entity include the early age of onset, the bone pattern, the high tendency to recurrence and the aggressive local behavior (8).

This case report highlights a JOF occurring in the mandibular anterior region which has made it a rare entity.

Case Report

In October 2010, a 6-year-old boy was referred to the Department of Oral Diseases of Mashhad Faculty of Dentistrywith a painless, progressiveswelling of the mandibular anterior region.According to his parents, he had the swellingfor 6 months but its size had increased rapidly during the last month. The patient had not experienced any pain or discomfort and no previous treatment had beencarried out for him.

He was otherwise fit and healthy. Extraoral examination revealed a moderately large left mandibular mass,causingasymmetry in the chin. No enlarged lymph nodes were found on palpation (Fig.1).

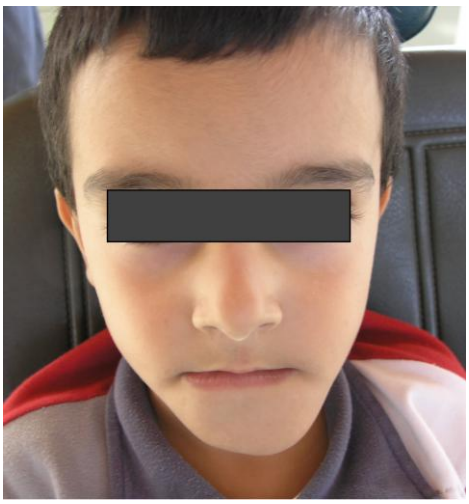

Figure 1. A 6-year old boy with a painless, progressive swelling of the mandibular anterior region which has caused an asymmetry in the chin

Intraoral examination showed expansion of the buccal and lingual cortical plates of the mandible, extending from left to right deciduous second molars.Displacement and mobility of mandibular incisors and deciduous canines were also observed. The mass was bony-hard and non-tender to palpation and overlying mucosa was smooth and normal in color.

The possibility of superimpositions in panoramic view in mandibular anterior region leads us to prescribe cone beam computed tomography (CBCT) instead.

CBCT can be used as a dose-sparing technique compared with alternative standard medical CT scans for common imaging tasks of oral and maxillofacial lesions.

CBCT sections showed a mixed, bilocular, well-defined lesion that contains dense, amorphous and odontome-like calcifications. A radiolucent rim was also seen around the lesion, which was extended from the mesial side of the unerupted left first mandibular molar to the mesial of the unerupted right first premolar (Fig. 2a-c). Expansion of the buccal and lingual plates was remarkable but the cortices were intact.

Gross displacement of mandibular incisors was obvious and unerupted canines were pushed towards the inferior border of the mandible.

Clinical findings such as rapid expansion, early age of onset, teeth displacement and midline location were indicative of aggressive central giant cell granuloma and mesenchymal malignancies, which were radiographically ruled out.

Other radiographic differential diagnoses were juvenile cemento-ossifying fibroma, calcifying odontogenic cyst (COC) associated with odontoma and ameloblastic fibro-odontoma.

Histologic examination of the non-capsulated lesion revealed benign neoplastic mesenchymal cells proliferation, which form a connective tissue stroma containing calcifications. The fibrous tissue was cell-rich in most parts and fibrillar in others and shows areas of hemorrhage and multinucleated giant cells as well. The mineralized component contains concentric lamellated ossicles and cementicles in various shapes, with peripheral eosinophilic rings around basophilic centers. Histologic examination confirmed the diagnosis of psammamatoid form of juvenile ossifying fibroma (Fig. 3).

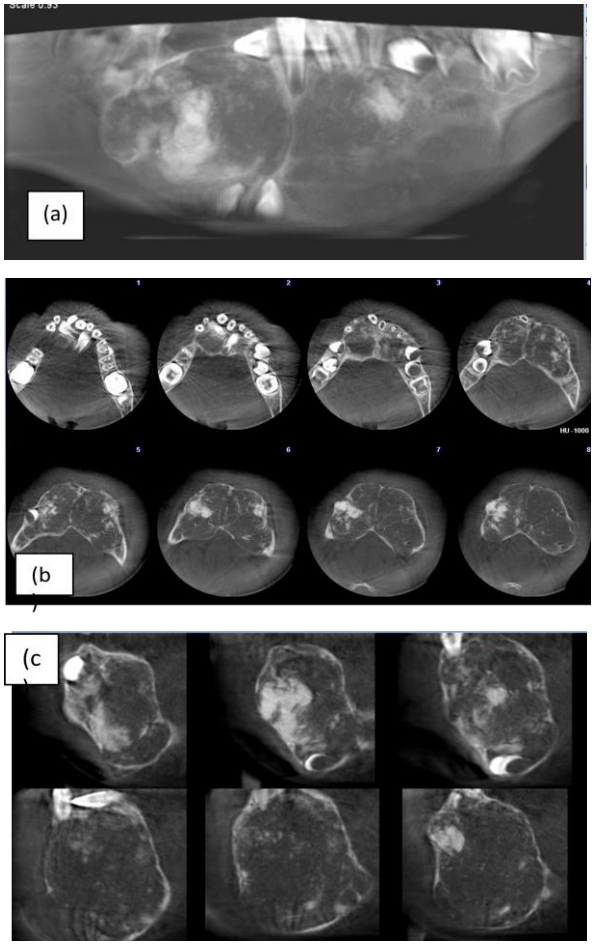

Figure 2. CBCT views of the lesion that shows the cortical expansion and the odontome-like calcifications in the lesion.
(2a) panoramic-like view. (2b) axial view.
(2c) cross sectional view

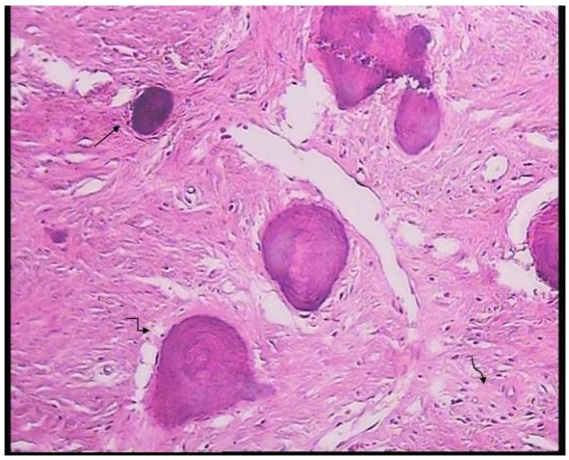

Figure 3. H&E photomicrograph of the tumor. shows cellular part of connective tissue. shows concentric lamellated ossicle with peripheral osteoid rim and pseudocystic degeneration. shows spherule of basophilic cementum-like material (original magnification x100)

Discussion

JOF has a predilection for the premolar and molar regions in both jaws (5) in contrast to our case in which it occurred in anterior mandibular region,crossing the midline.

Radiographically, juvenile COF resembles conventional COF but due to its distinct histological features,the most common site of involvement, clinical behavior (aggressiveness and recurrences)and also the age of occurrence, it has been considered as a separate entity and classified as a clinicopathological variant of the ossifying fibroma (10,11).

JOF typically appears as a slow-growing asymptomatic enlargement. Some lesions, however, demonstrate rapid growth and may be associated with local complications as it achieves a large size and exhibit aggressive behavior (12,13).

At first glance, rapid expansion in our case may seemslike a malignancy but teeth displacement without mobility and resorption as well as intact cortical plates makes it closer to benign lesions.

JOF is not encapsulated. Its periphery is well-defined and corticated though. It appears locally aggressive, with cortical disruption and involvement of many adjacent anatomical structures. This lesion has predominating soft tissue consistency with variable amounts of internal calcifications and/or linear or irregular focal bone (4).

In our case, the lesion is well-defined and surrounded with a radiolucent rim, which shows the central maturation of the lesion.The lesion is mixed and contains odontome-like calcifications.

Given the radiographic appearance and the patient's age, differential diagnoses include Juvenile cemento-ossifying fibroma, COC (calcifying odontogenic cyst associated with odontoma) and ameloblastic fibro-odontoma.

The JOFs are classified into two different clinicopathological groups: the trabecular and the psammomatoid types. Trabecular JOF is recognized by the presence of trabeculae of fibrillar osteoid and woven bone. Psammomatoid JOF is characterized by the presence of small uniform spherical ossicles, called psammoma bodies. The most reported sites of these bodiesare sinonasal and orbital cavities (14). In the present case, histopathologic findings were similar topsammomatoid type of JOF, which isa rarity in the anterior region of the mandible.

The undulating expansion which is also found in this caseisunusual for JCOF, as the JCOF expansion is more concentric.

An uncommon clinical entity, JOF is an aggressive lesion mimicking malignancy similar to osteosarcoma. Therefore, it is of paramount significance for it to be

diagnosed as soon as possible so that it can be treated accordingly (9).

Conventional treatment for JOF includes total excision due to its high recurrence rate (30-56%) (15).

Mandibular resection was performed for this patient from mesial of the first permanent molar on the left side to the mesial of first permanent molar on the right side.The patient was treated and underwent followup.

Conclusion

It is essential to make the right diagnosis at the right time,for the best treatment,and following up the patient accordingly.

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