Coincidence of Compound Odontoma and Cemento Ossifying Fibroma; A Rare Case Report

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Abstract

Objective: Cemento-ossifying fibroma defines as a relative rare osteogenic neoplasm of the jaw. This tumor includes fibrous and osseous components. Odontoma is the most common odontogenic tumor containing enamel ,dentin ,cementum and pulp tissue. in this paper we report a rare case of ossifying fibroma associated with compound odontoma in the mandible**.**

Case: A 37-years-old woman was referred to Oral Medicine department , Shahid Beheshti Dental School with complaint of swelling in the anterior part of the mandible, over 6 years period. Clinical examination revealed mandibular enlargement in right – anterior region with labial and lingual expansion and canine missing . panoramic view showed a large mixed radiolucent - radiopaque lesion associated with impacted canine.The differential diagnosis include calcifying odontogenic cyst (COC) and cemento-ossifying fibroma(COF). Histopathologic examination established diagnosis of COF with multiple compound odontoma .

Conclusion: The relationship between the occurrence of these two lesions is not clear and more studies are needed to establish the relationship between them.

Key words: Fibroma, Ossifying; Mandible; Odontoma; Odontogenic Tumors.

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Introduction

Fibro-osseous lesions (FOLs) in the craniofacial bones consist of a group of lesions, which are composed of hypercellular fibrous and osseous elements, both exhibiting a wide spectrum of variations. The majority of FOLs have overlapping histopathological features; thus, accurate diagnosis requires a thorough history, clinical and radiographic examinations and analysis of histological features. The FOLs of the maxillofacial region include fibrous dysplasia, benign fibro-osseous neoplasms and reactive (dysplastic) lesions (1,2). Cemento-ossifyng fibroma is a subtype of FOLs, in which the normal bone structure is replaced with fibroblasts, collagen (3,4) and mineralized tissue with variable degrees of mineralization including woven bone or cementum-like masses (5). Cemento-ossifyng fibroma is a benign asymptomatic slow-growing tumor, which is most frequent in females with a peak incidence in the third and fourth decades of life (2,6). It is believed to originate from the periodontal ligament but similar neoplasms have also been reported in other craniofacial bones, which questions this theory (2,4).

Odontomas are the most frequent

odontogenic tumors originating from the odontogenic epithelial and mesenchymal tissues (2). There are several etiologic theories, which include local trauma, infection, gene mutations or possible postnatal interference with the genetic control of tooth development (7). Complex odontomas refer to the haphazard arrangement of tooth elements such as enamel matrix, enamel, tubular dentin and pulpal tissue. Compound odontomas refer to the aggregation of recognizable teeth. Both types of odontomas are primarily diagnosed in children (8). Odontoma grows slowly and painlessly and may occur in tooth-bearing areas of the jaws.

A few articles have reported simultaneous occurrence of two or more odontogenic tumors or central lesions in the jaws (9,10).There is one report of subsequent occurrence of COF at the site of odontoma (2) and another article describing complex odontoma associated with ossifying fibroma (4). Herein, we report uncommon occurrence of multiple odontoma and COF in the same region.

Case:

A 37 years-old woman was referred to Oral Medicine Department, Shahid Beheshti dental school with complaint of swelling in the anterior part of the mandible, over 6 years long .She has facial asymmetry without lymphadenopathy.

There was an expansion in the right side of anterior region of mandible, extending from the right first premolar to the central incisor in the left side. Missing of canine and slight displacement of the first premolar and second incisor was seen. There was intact Overlying mucosa, no mobility on palpation, tenderness or paresthesia. Vitality test was performed on the first premolar, first and second incisor and they were vital. Panoramic view revealed a mixed radiolucent-radiopaque lesion located in the right anterior region of mandible from second premolar to the left first incisor, and an impacted canine was seen inside the lesion. An alteration of mandibular bone structure at the level of inferior cortex and downward expansion was seen (Figure1).

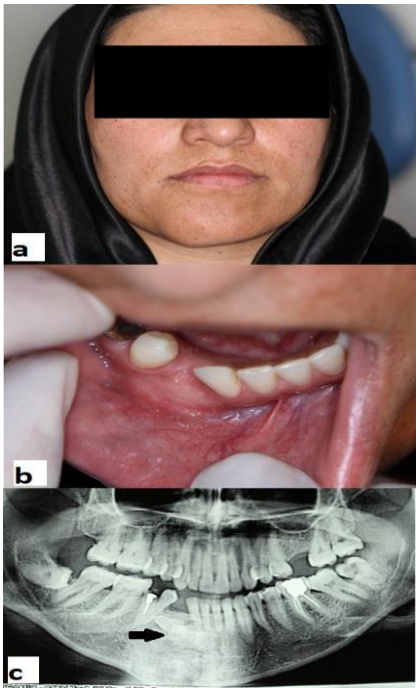

Figure 1- *a* .asymmetry on the face *b*. missing of canine, expansion of labial cortex and displacement of the right second incisor *c*.Panoramic view showing a large well circumscribed radiolucent- radiopaque lesion in the right anterior region of the mandible. Inferior cortical expansion, impacted canine (arrow) and displacement of first premolar and second incisor are seen

In Cone beam computed tomography (CBCT) a mixed radiolucent-radiopaque lesion with multiple tooth like structure and impacted canine was seen. First premolar

and second incisor showed root divergency. there was no evidence of root resorption. In the sagital view the areas of cortex perforation were detected (Figure2).

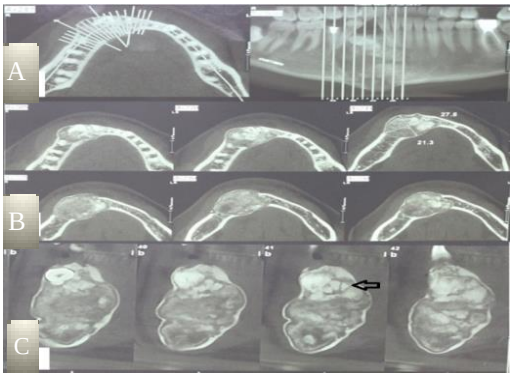

Figure 2- CBCT imaging, axial view shows a lesion with bucal and lingual expansion and multiple radiopaque internal structures.(A) In this images impacted canine and multiple tooth –like structures are seen that surrounded by less radiopaqe structures.(B) CBCT imaging , Sagital view. sections of impacted canine, tooth –like structures (arrow) and radiopaque elements(C)

Our differential diagnosis, on the basis of clinical and radiographic examination were: calcifying odontogenic cyst (or Gorlin's cyst) which usually develops in the incisor-canine regions and radiographically presents as a well-circumscribed, unilacular radiolucency that contains irregular calcifications, additionally it is sometimes associated with unerupted teeth or odontoma (8) and Cemento-ossifying fibroma (COF) that shows a predilection for females. The mandible is the most commonly involved site. The radiographic borders appear well defined and mostly corticated. The density of the lesion is mixed. The lesion tends to be concentric within the medullary part of the bone with outward expansion approximately equal in all directions (6).

Surgical excision was done without primary incisional biopsy by surgeon decision in the basis of patient's history and clinical and radiographic examination. The lesion, including the impacted canine were compeletly excised. Extraction of First and second premolars was done. Empty bone cavity curettage was done and defect was filled with allogenic bone. Patient's tolerance and cooperation was good.

Microscopic findings revealed a fibro-osseous lesion composed of cellular fibrosis connective tissue with spherules and large globular structure of calcified material. In the lesion cementum-like and osteoid like material concentrated and form large irregular bony tissue. Hemorrhage in periphery of the lesion was seen. The other part of the lesion was composed of tooth like structures with tubular dentin around the pulp-like tissue. Excisional biopsy of the lesion presents a cement-ossifying fibroma associated with multiple compound odontomas (Figure 3).

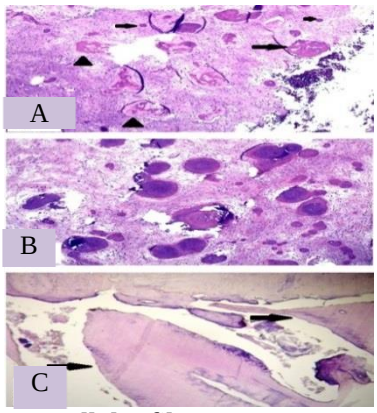

Figure 3- cellular fibrous stroma consist of calcified materials , globes of cementum (arrow) and bone (arrow head) with osteoids and osteoblastic rims. 100× (A). fibroblasts proliferation with globes of cementum and giant cells are seen. 200 ×(B) two small tooth structures(arrow) contains of dentin pulp and enamel matrix. 100 ×(C)

The diagnoses were confirmed based on excisional biopsy specimens. No recurrences

or post-operative complications were observed during the 6 months follow-up period and panoramic view showed healing of lesion (Figure 4).

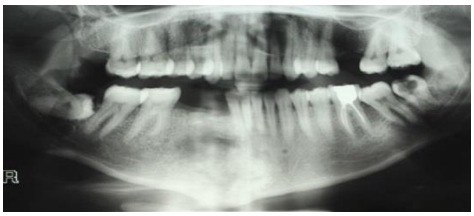

Figure 4- Panoramic view after 6 month follow up, site of surgery shows healing

Discussion

In 1971 the World Health Organization (WHO) classified four types of cementum-containing lesions: fibrous dysplasia, ossifying fibroma, cementifying fibroma and cemento-ossifying fibroma.According to the second WHO classification, benign fibro-osseous lesions in the oral and maxillofacial regions were divided into two categories, osteogenic neoplasm and non-neoplastic bone lesions; cementifying ossifying fibroma belonged to the former category. However, the term ‘‘cementifying ossifying fibroma’’ was reduced to ossifying fibroma in the new WHO classification in 2005 (6). Ossifying fibroma of craniofacial bones is a benign neoplasm mainly composed of two components: fibrous stroma and elements that show various degrees of maturation. The stroma consists of varying amount of fibroblasts and loose to dense fibrous tissue. Calcified elements include mineralized bodies (ossicles), osteoids, fiber bone (woven bone), and mature bone(lamellar bone). Globular cementum-like masses may be present either alone or together with the trabeculae (1).

Cement-ossifying fibroma is a true neoplasm with a significant growth potential .This tumor is more frequent in the premolar and molar regions of the mandible in the third and fourth decades. Female are more commonly affected. Clinical presentation is usually a spherical or ovoid expansion of the jaw. Small lesions are rarely symptomatic and usually are discovered during radiographic examinations. Larger lesions cause painless enlargement of the involved bone and Considerable facial asymmetry .Pain and paresthesia are rarely associated with COF (11).

Radiographically a well-defined and unilocular Radiolucent-radiopaque lesion is seen, depending on the amount of calcified material production. Root resorption or divergency of teeth may be observed. Large COF of the mandible often demonstrate a characteristic down ward bowing of the inferior cortex (4). Compound odontoma occurs most commonly in the anterior maxilla and consists of a collection of numerous small teeth (12).

Although the lesion is non-aggressive, occasionally they can cause obvious asymmetry of jaw (4).

In this case, patient reports a history of asymptomatic and long term growing of the lesion on her jaw, with bony hard consistency which suggest, a nonaggressive intrabony lesion. occurrence of simultaneous odontogenic lesions or simultaneous odontogenic and non-odontogenic lesions, described combined lesions, sometimes called hybrid lesions which is extremely uncommon and have been reported in a few papers(2,4).

This patient had an impacted canine, compound odontoma and cemento-ossifying fibroma. The origin of COF in this case is not identified, although earlier age of development of odontoma could suggest that COF is derived from odontomas.There is only three reports similar to our case until now; Matsuo K. et al. (2) reported multiple complex odontomas and subsequent occurrence of an ossifying fibroma in a 3 year-old boy; In this report the patient was followed for years and OF occurred after odontoma. Due to this report, the de novo occurrence of OF in the overlapping area of adontoma or origination from pluripotent cells from the remnants of the complex adontoma is suggestive, in our case COF and odomtoma explored in same time and the patient had no previous imaging so the time of tumors formation is not justifiable, although odontomas occurs in earlier age than COF (12).

Similar previous report, Ohtake K. et al. (13) described three complex odontomas associated with ossifying fibroma(OF) in a 10 year-old boy; author suggested that under certain conditions , an immature element of an odontoma near the root of an existing tooth may develop into an ossifying fibroma. Agha Hosseini F. et al. (4) reported concurrent occurrence of cement-ossifying fibroma, periapical cement-osseous dysplasia and complex odontoma in a 46 years-old woman for the first time.the auther emphasized more case reports are needed to determine the relationship of this coincidence.

There are some papers which reports simultaneous occurrence or occurrence in different time of two or more central odontogenic lesions in the jaw (2,13).It suggests the existence of a local environmental condition such as trauma, growth factor, that permits the growth of multiple odontogenic lesions.

Also occurance of two lesion developed close to each other in the fairly limited area of alveolar bone suggests the possibility of a local environment permissive for the growth of multiple odontogenic tumors. It would be proposed to research the gene expression in this local environment and the genetic background of patients with like lesions (4).

Conclusion

The relationship between the occurrence of these two lesions is not clear and more studies are needed to establish the relationship between them.

Acknowledgment

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Conflict of Interest: "None Declared"

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