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Case Report

A Rare Case of Juvenile Cemento-Ossifying Fibroma: Diagonosed by Cytological and Radiological Correlation

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Abstract

Cemeto-ossifying fibroma (COF) is a benign fibro-osseous lesion of the mandible and maxilla. The World Health Organization (WHO) introduced the term "cemento-ossifying fibroma" by combining two entities, cementifying fibroma and ossifying fibroma in 1992. However in 2005 the entity was included under ossifying fibroma group. The tumor usually affects the young adults. Rarely, this tumor arises in children, known as Juvenile aggressive ossifying fibroma, having a more aggressive clinical course. They arise in the mandible in 62% to 89% of patients, majority occur in the premolar region but rare in maxilla. But the juvenile variant commonly involves maxilla. They are thought to arise from the periodontal ligament and are composed of varying amounts of cementum, bone, and fibrous tissue. Herein we are presenting a rare case of juvenile cemento-ossifying fibroma, arising from maxilla in a 12 years old girl diagnosed by radiological and cytological correlation and confirmed by histopathological examination.

INTRODUCTION

Cemento-ossifying fibroma (COF) is considered a benign fibro osseous lesion (FOL) [1]. The World Health Organization (WHO) introduced the term "cemento-ossifying fibroma" by combining two entities, cementifying fibroma and ossifying fibroma in 1992. However in 2005 the entity was included under ossifying fibroma group [1,2]. COF is subclassified based on age as juvenile COF(JCOF) and conventional COF. JCOF is locally aggressive [5]. The most common location of COF is the mandible comprising 70-90% of all cases. [8] Whereas JCOF usually involves the maxilla and paranasal sinus. [5] Although central COFs of the mandible are common, central COFs of the maxillary sinus are not; a few have been reported in literature. Here we are presenting a rare case of juvenile cemento-ossifying fibroma arising from maxilla suspected preoperatively by cytological examination, later confirmed by histology.

CASE PRESENTATION

A 12 years old girl presented with gradually increasing hard mass in right half of face for last two years with relatively rapid growth for last four months. There was obvious facial deformity with right nostril obstruction. (Figure 1a). In CT scan mass appears to arise from maxillary sinus, (8 X 6) cm^2 in size, having calcified septation with cystic spaces. (Figure 1b and 1c). Fine needle aspiration cytology (FNAC) was done from relatively soft part. The smears were hypo-cellular, composed of scattered osteoid material, few small clusters of benign spindle cells, occasional benign osteoblast and few giant cells. No malignant cell was found, however occasional regenerative multi-nucleated muscle cells (caterpillar cell) were detected indicating probable muscle degeneration and regeneration by compression effect (Figure 2). A presumptive cytological diagnosis of benign fibro-ossious lesion, probably JCOF was rendered considering the clinical as well as radiological features.

The preoperative diagnosis was confirmed by subsequent histopathological examination of incisional biopsy specimen. The section showed proliferation of benign fibroblasts and formation of osteoid material, rimmed by osteoblasts in some areas. There were few basophilic structures, having concentric lamellar calcification simulating psammoma bodies (Figure 3a and 3b). Thus, final diagnosis of JCOF or Juvenile Psammamatoid Cemento- Ossifying fibroma was rendered. Lesion was excised by hemi-maxillectomy. Post-operative recovery was uneventful, and patient was discharged after 1 week following surgery.

DISCUSSION

Common to all FOLs is the replacement of normal bone by a tissue composed of collagen fiber and fibroblasts that contain varying amounts of mineralized substance, which may be bony or cementum-like in appearance.[3] According to recent WHO classification of FOLs, Juvenile trabecular OF and juvenile psammamatoid OF are two histologic variants of OF. Due to the presence of cementum-like material, OF have been called as cementifying fibroma (CF) and COF, if they have both cementum and bone-like material [4]. The JCOF is a benign but locally aggressive lesion that invades and destroys tissues until the eyes and the cerebrospinal space are affected [5,6]. It originates from the mesenchymal blast cells of the periodontal ligament, and with a potential to form fibrous tissue, cement and bone, or a combination of such elements. However, there is controversy over such an origin, since tumors of similar histology have been reported in bone lacking periodontal ligament, such as the long bones of the body [7].

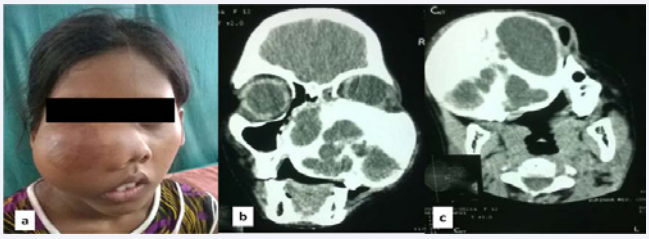

Figure 1 (a): Clinically a huge right Maxillary mass causing facial deformity (b) and (c) : CT scan showing well defined mass with calcification and soft tissue attenuation.

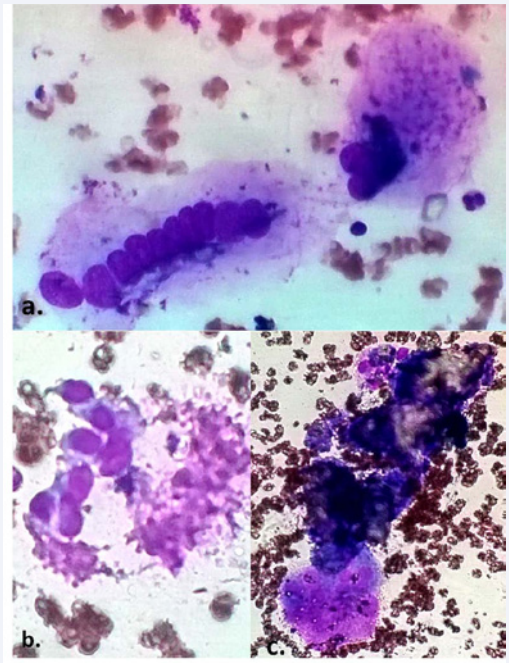

Figure 2 Cytological findings of the mass lesion. (a) Isolated multinucleater giant cells (b) Scattered benign spindle cells. (c) Osteoid material along with a giant cell.

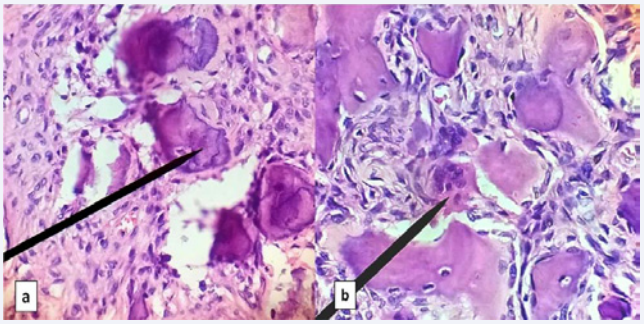

Figure 3 Histopathological findinds (H and E Stain, 40X) (a) Proliferation of benign fibrous tissue with formation of osteiod and cementum like material. Psammamatoid body (pointer). (b) Presence of giant cell (pointer) and osteoblastic rimming of osteoid material.

COF is most commonly seen between the third and fourth decades of life and is more frequent in women than in men (4:1) [2]. The most common location is the mandible, which comprises 70-90% of all cases. The facial sinuses and the nasal cavities are rarer locations [8]. Whereas JCOF mostly involves the maxilla and paranasal sinus [5]. Clinically, these tumors manifest as a slow-growing intra-medullary mass that is normally well delineated and asymptomatic, though over time the lesion may become large enough to cause facial deformation [2]. It is more or less firm in consistency, depending on its degree of mineralization. The COF is covered with normal mucosa [8]. Radiologically, COFs are usually well circumscribed masses which expand the underlying bone. They are usually small, but can become large, particularly when they arise from the maxilla or paranasal sinuses. They are initially radio-lucent on x-ray with soft tissue attenuation on CT, increasing amount of calcification or ossification is seen with maturation. They usually expand the bone without cortical breach [9]. There are few similar entities that occur in maxillofacial region, they can be distinguished from COF by careful radiologic evaluation. In contrast to fibrous dysplasia, which has a blending margin with the surrounding bone, COF produces round well defined tumor mass. Cemento-osseous dysplasia shows the presence of bony cysts with a wide sclerotic border and it is multifocal while COF is not. Pindborgs tumor may be associated with impacted teeth and shows an appearance of driven snow in the radiograph. Odontoma shows presence of tooth-like structures whereas COF shows a radio-opaque focus [10].

Cytology smears are usually paucicellular composed of clusters of oval and spindle-shaped fibroblasts with no atypical features in haemorrhagic background [11]. In the present case the spindle cells were admixed with spherical calcified structures. Few giant cells and regenerative muscle cells were also noted. In histopathology, it shows irregular calcifications within a hypercellular fibrous connective tissue stroma. The calcifications are extremely variable in appearance and represent various stages of bone and cementum deposition[12]. Cementifying fibroma produces compact basophilic nodules, whereas ossifying fibroma has trabeculae of osteoblasts pitted with osteocyte cavities.COF shows combination of both. It differs from fibrous dysplasia by presence of osteoblastic rimming around osteoid material [3] JCOF possess two different types of morphologic features one is psammomatoid another is trabecular. Psammomatoid COF has

ovoid ossicles while the trabecular COF has cellular or acellular trabecular bones [5].

JCOF can be distinguished from adult type by its clinical, radiological, and pathological characteristics. Because of its aggressiveness, it is often mistaken for osteosarcoma, osteoblastoma, or other malignant bone tumors. Thus, it is crucial to distinguish it from malignant bone tumors or other FOLs [5]. The aetiology of JCOF has not been fully understood. Genetic features in FOL of craniofacial bone are not well established, but there are reports that COFs were related to non-random break points at Xq26 and 2q33. The treatment of choice for COF is complete surgical resection and it is usually cured by surgery. Incomplete resection of the tumor may result in recurrence [5]. In conclusion, the diagnosis and typing of cemento-ossifying fibroma is obtained by evaluating clinical and radiological features of the lesion. Classical radiological picture along with presence of benign fibroblasts, calcified material, osteoblasts and multinucleated giant cells in cytology can provide a proper preoperative diagnosis.

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