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DIAGNOSTIC DILEMMA IN JUVENILE OSSIFYING FIBROMA OF MANDIBLE – A CASE REPORT

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ABSTRACT Juvenile ossifying fibroma (JOF) is an uncommon, benign, bone-forming neoplasm that is distinguished from other fibro-osseous lesions primarily by its age of onset, clinical presentation, and potential behavior. This lesion most often occurs between 5 and 15 years of age, shows a male predilection, and may exhibit rapid growth of the involved anatomic site, sometimes resulting in considerable facial disfigurement.Most juvenile ossifying fibromas arise in the vicinity of the paranasal sinuses. With regard to the incidence of JOF in the jaws, there are conflicting reports of maxillary and mandibular predilections. This report details the diagnosis and treatment of a 12-year-old boy presenting with a mandibular swelling that was subsequently determined to be JOF.

KEYWORDS : Juvenile ossifying fibroma, tumors of mandible, fibroosseous Lesions.

INTRODUCTION

Maxillofacial fibrosseous lesions usually present a diagnostic dilemma for clinicians and pathologists. According to the WHO classification, the term juvenile ossifying fibroma (JOF) is used for an actively growing lesion mainly affecting persons younger than age 15.

JOF is an uncommon, benign, bone-forming neoplasm that is distinguished from other fibro- osseous lesions primarily by its age of onset, clinical presentation, and potential behavior. This lesion most often occurs between 5 and 15 years of age, shows a male predilection, and may exhibit rapid growth of the involved anatomic site, sometimes resulting in considerable facial disfigurement. Histologically, it consists of a cell-rich fibrous stroma, containing bands of cellular osteoid without osteoblastic rimming, together with trabeculae of more typical woven bone. Small foci of giant cells may be present. The lesion is non-encapsulated but well demarcated from surrounding bone. It is usually asymptomatic, achieving a large size and exhibiting aggressive behaviour, and is often diagnosed as 'juvenile ossifying fibroma', 'aggressive ossifying fibroma' or 'active ossifying fibroma' in the literature. This report details the difficulty encountered in diagnosis and formulation of treatment plan of a twelve year old boy presenting with a mandibular swelling that was subsequently determined to be JOF.

CASE REPORT

A twelve year old boy was referred to the Department of Oral and Maxillofacial Surgery at the Father Muller's Charitable Institution , Mangalore, Karnataka , India. The child presented with a swelling in right lower jaw associated with pain since three months. The lesion had been slowly increasing in size since it was first noticed. There was no history of trauma .

Pain was dull-aching , intermittent and relieved by the pain-killers. Physical examination showed a healthy, normally developed young boy in no apparent distress. There was a detectable facial asymmetry caused by an approximately 3 × 3-cm mass involving the right mandibular angle and ramus area . No bruits or pulsations were detected.The mass was firm and nontender to palpation and not adherent to the overlying skin. Mouth –opening was restricted since two weeks . On intra-oral examination, a swelling was present distal to right second permanent molar till retromolar region on right side of mandible. There was expansion of the right buccal cortex which was firm to palpation. Significant lingual expansion of the inferior cortical bone also was noted. There was no evidence of tooth mobility, abscess formation, dehiscence or malocclusion. On examination of the neck,single right submandibular lymph node was palpable which was firm, nontender, 1.5 x 1.5 cm in size and mobile.

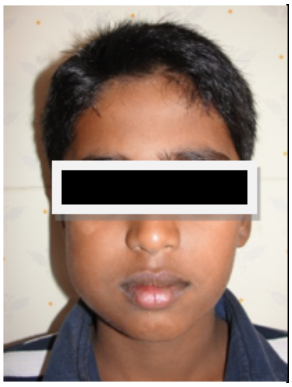

Figure 1-extraoral swelling

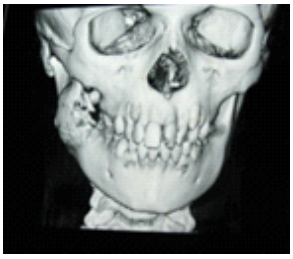

Figure 2- lytic lesion in mandible

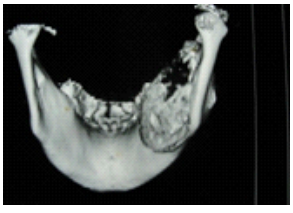

Figure 3- destruction of lingual cortex

Pre-operative

Radiologic examination revealed an unilocular radioluscency of the right mandibular body and ramus region distal to right second permanent molar with an unerupted molar seen within the lesion on OPG X-ray and lateral oblique mandible X-ray. Computed tomography (CT) showed an expansile lytic lesion

involving the right ramus, angle and body of mandible with laminated periosteal reaction . The lingual cortex seemed to be perforated.

A needle aspiration of the involved area performed at the time of initial presentation did not yield fluid. An incisional biopsy was subsequently performed, providing a diagnosis of aggressive osteoblastoma or osteosarcoma. Finally, a diagnosis of odontogenic tumor (osteoblastoma) was made on the basis of the clinical and radiographic findings.

Complete excision of relatively solid tumor mass was performed under general anesthesia through the submandibular approach . The lateral cortex was thinned, but not perforated, by the tumor. However, the lingual cortex was perforated. The right submandibular lymph node was also excised along with submandibular gland. The inferior alveolar nerve was preserved and the mandible was stabilized with a 2.5-mm 14-hole reconstruction plate. Hemostasis achieved and suction drains placed before closure of wound in layers. Post – operative stay of patient was uneventful and the patient was discharged on the tenth day after the surgery.

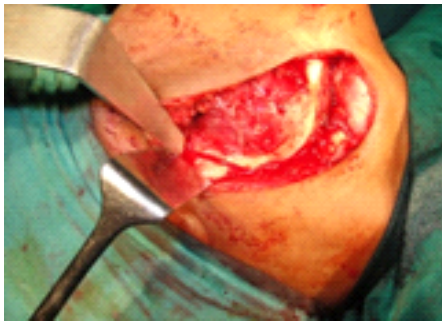

Figure 4-exposure of tumor site

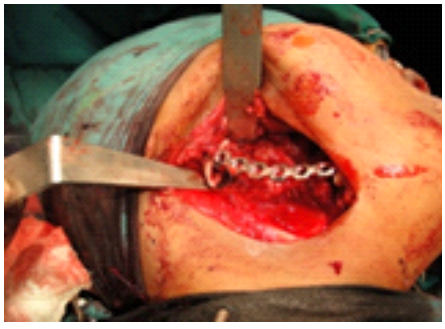

Figure 5-excised tumor and reconstruction plate

Intra-operative

Microscopically, the excised specimen consisted of spindle cells arranged in storiform pattern.

Amidst these, there were bony trabeculae lined by osteoblasts. These was no evidence of cellular atypia, mitoses or necrosis. The histology of the specimens was typical for juvenile ossifying fibroma.

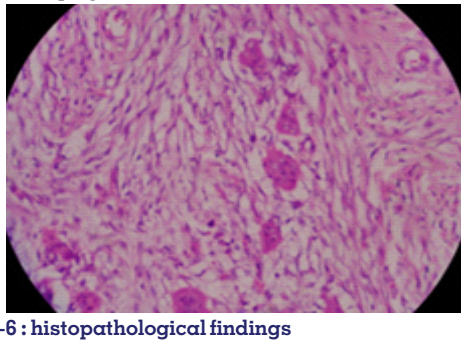

Figure-6 : histopathological findings

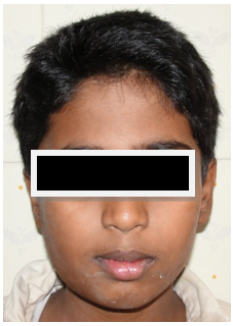

Figure 7- 7th post-op day

Post-operative

DISCUSSION

Juvenile active ossifying fibromas are true fibro-osseous neoplasms. JOF may present clinically as either a gradual or rapid , painless expansion of the affected bone or region. Nasal obstruction, exophthalmos, and, rarely, intracranial extension can be associated with those lesions arising within the paranasal sinuses and orbit . As the term " juvenile " emphasizes, the tumor occurs mainly in children with 79% of them being 15 years old or younger.

The term "active" describes the tendency for a local aggressive growth pattern rather than for rapid growth. In general, juvenile ossifying fibroma has a more aggressive growth pattern than the ossifying fibroma, which is encountered in the third and fourth decades of life. In our case also, we observed aggressive nature of JAOF as the lesion grew double its size after the incisional biopsy was taken.

Radiographic features are nonspecific and typically consist of a unilocular or multilocular radiolucency having ill-defined borders and occasional central opacification. Aggressive lesions may show cortical thinning and perforation which was also seen in our case.

Histologically, the typical features of JAOF are a densely cellular stroma containing calcified components. The degree of maturation of the calcification can vary, but the peripheral osteoid rims surrounding the mineral components are an important feature in JAOF. Although the tumor tissue is usually characterized by a relatively fibrotic, avascular stroma, moderate vascularization has also been reported. The highly cellular nature of the fibrous matrix and woven bone reflects the more aggressive behaviour of the tumor which was similar to our case . There is some degree of histologic overlap between JOF and cemento-ossifying fibroma.

In contrast to cemento- ossifying fibroma, which histologically is characterized by uniformity of pattern, JOF is more likely to show markedly heterogeneous morphology , a characteristic that may complicate the diagnosis and subsequent management of these tumors.

The rapid growth rate often exhibited by these lesions can be quite alarming and cause the clinician to suspect the presence of a malignancy. Malignant neoplasms that have a tendency to involve the craniofacial skeleton in this age group include osteosarcoma, chondrosarcoma, Ewing's sarcoma, and the African form of Burkitt's lymphoma. Benign neoplastic or developmental lesions are more commonly encountered in the jaws, and, although they may show differing degrees of aggressiveness, they often present with the radiographic characteristics of JOF. Included among these diseases are osteoblastoma, an intraesseous form of fibromatosis known as desmoplastic fibroma, cemento-ossifying fibroma, fibrous dysplasia, central giant cell granuloma, and cherubism. Also occurring in the jaws are the benign odontogenic neoplasms

and cysts, which include, in part, ameloblastoma, ameloblastic fibroma, ameloblastic fibro- odontoma, adenomatoid odontogenic tumor, and calcifying odontogenic cyst.

Since, fibro-osseous lesions of the maxillofacial complex especially JOF are often difficult to diagnose from both a clinical and a histopathologic point of view, the differential diagnosis with other fibro-bony lesions are mainly determined by the characteristics of the calcified tissues. Owing to of this lesion's aggressive nature and high recurrence rate, early detection and complete surgical excision are essential.

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