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Case Report

A rare exophytic juvenile ossifying fibroma mimicking an extraosseous lesion: a diagnostic challenge

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ABSTRACT

Juvenile ossifying fibroma (JOF) is a rare fibro-osseous lesion that predominantly affects children and adolescents and may present a diagnostic challenge due to its variable clinical and radiologic features. We report a rare case of a 9-year-old boy with an atypical presentation of JOF in the mandible, clinically mimicking an extraosseous lesion. Conventional imaging was inconclusive, whereas CT demonstrated a well-defined exophytic lesion with a hypodense centre and hyperdense peripheral margin. Histopathological examination confirmed the diagnosis of JOF. This case underscores the diagnostic difficulty associated with this underreported and unusual presentation.

Keywords: Juvenile ossifying fibroma, Fibro-osseous lesion, Computed tomography, Exophytic lesion, Paediatric mandibular lesion

INTRODUCTION

Ossifying fibromas are benign fibro-osseous growths located in the jaw and craniofacial area.[1] The WHO characterizes juvenile aggressive ossifying fibroma as a rapidly growing lesion, predominantly affecting individuals younger than 15 years, composed of a fibrous stroma rich in cellular osteoid and lacking osteoblastic rimming.[2] This neoplasm is extremely rare and is a subject of great debate amongst experts, especially in differentiating it from its adult variant based on age, site, behaviour and microscopic features.[3] It is commonly seen in the first and second decades of life.

While JOF usually presents as a well-defined intraosseous mixed lesion, this case showed atypical features in an uncommon site, mimicking an extraosseous lesion, thereby highlighting the diagnostic challenge and importance of advanced imaging.

CASE REPORT

A 9-year-old boy reported to the Department of Oral Medicine and Radiology with a one-month history of swelling in the posterior lower right cheek region, which had become tender over the past week. The swelling gradually increased to present size. He had a history of fever about 1.5 months ago, with no associated trauma or pain. Extraoral examination showed facial asymmetry and a diffuse swelling of approx. 1×1 cm in the right mandibular ramus angle region (Figure 1). The overlying skin was normal. On palpation, swelling of approx. 0.5×0.5 cm which was firm to rubbery, tender and fixed to the inferior border of mandible was palpated. Multiple lymph nodes were palpable, with mobile and tender right submandibular lymph nodes. Intraoral examination revealed vestibular tenderness w.r.t 46, with no other significant findings. A provisional diagnosis of tubercular lymphadenopathy was made.

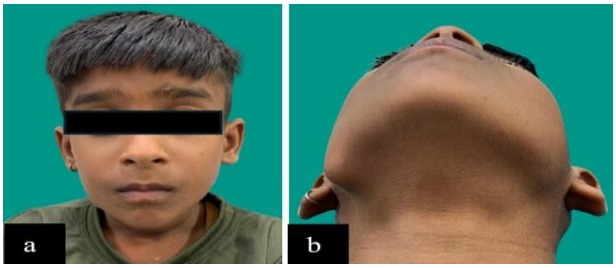

Figure 1 (a and b): Extraoral pictures.

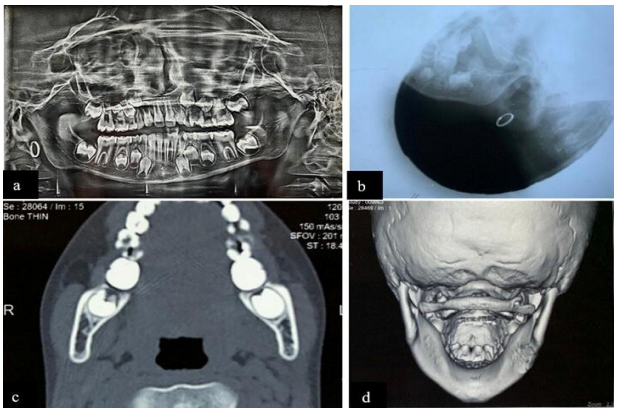

Figure 2: (a) Mild marginal irregularity in the right inferior border of mandible i.r.t 46 47 region. (b) Extraoral lateral oblique radiograph showing no significant finding. (c) CT image reveals exophytic, well-defined lesion with a hypodense center and hyperdense periphery on the lower and lateral border of right mandible i.r.t 46 47 region. (d) 3DCT image.

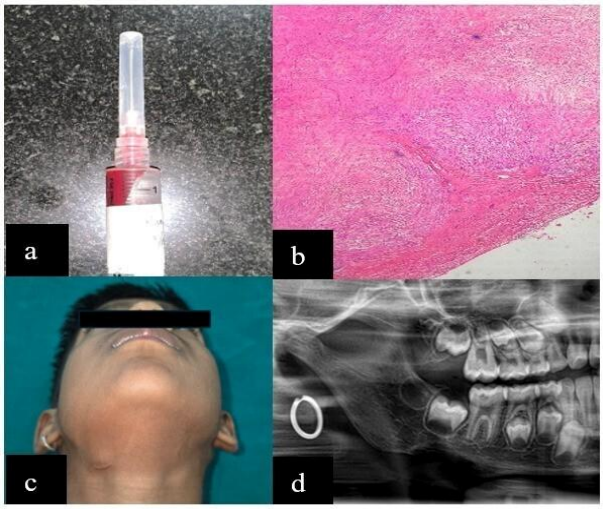

Figure 3: (a) FNAC yielded bloody aspirate. (b) Histopathological picture shows the background mainly comprised of plump proliferating fibroblasts with hyperchromatic nuclei and few mitotic figures. Cells are arranged in storiform pattern at some places intermixed with loosely arranged myxomatous components, cystic degeneration, haemorrhagic areas, osteoid components and mild response of chronic inflammatory cells. (c and d) 6 months follow-up.

Initial radiograph did not reveal any significant findings except for marginal irregularity of the inferior border of mandible in Orthopantomogram (Figure 2a, 2b). Ultrasound of the region revealed a cystic lesion in the right submandibular area. CT imaging demonstrated a clearly outlined exophytic lesion with a central hypodensity and denser outer margins along the right inferolateral border of mandible with expansion of adjacent cortex without bony erosion (Figure 2c). To rule out tuberculosis, FNAC (Figure 3a) and CBNAAT and Zn stain was done, which tested negative for Mycobacterium tuberculosis. Following, excisional biopsy was performed by submandibular incision under general anaesthesia and the final diagnosis was made as Juvenile Ossifying Fibroma confirmed by histopathology (Figure 3b). The patient has undergone 6 months follow-up (Figure 3c, 3d) and continues to be under follow-up for 1 year and then 3 years.

DISCUSSION

Ossifying fibroma is a benign lesion that progressively expands within the bone, leading to asymmetry, facial deformity, and malocclusion.[5] OF is a true neoplasm of mesenchymal origin, that likely originates in the cells of the periodontal ligament.[6,7]

Chromosomal aberrations and HRPT2 gene mutations have been implicated in ossifying fibromas, especially in hyperparathyroidism-jaw tumor syndrome.[6,8] Potential provoking factors include local trauma, tooth removal, periodontal disease, and congenital osseous maturation abnormalities. But the true nature of ossifying fibroma (OF) is still unknown and controversial.[6]

JTOF often presents without symptoms and may be detected incidentally during routine radiographic evaluation. Displacement of teeth may be the first sign of the tumour. JTOF has a potential for pseudo tumorous growth, and when it grows, it may grow rapidly to cause facial asymmetry and jaw deformation.[4] In the present case, mild facial swelling was noted.

Radiographically, JTOFs typically appear as unilateral, unilocular mixed radiolucent-radiopaque lesions with corticated borders, though they may be entirely radiolucent with faint opacities. CT is essential for assessing larger lesions. They expand concentrically, often displacing teeth and the inferior alveolar canal. Although significant expansion and thinning may occur, the outer cortical plate typically remains preserved.[4]

In this case, the lesion did not adhere to the typical presentation of juvenile trabecular ossifying fibroma (JTOF). OPG did not provide any key diagnostic features. CT scan demonstrated a well-defined exophytic lesion with hypodense centre and hyperdense peripheral margins, which is an atypical presentation of JTOF that may obscure its intraosseous origin.

Management strategies for JOF lack a universally accepted protocol, and vary widely often depending on lesion location, extent, and associated symptoms. Neurologically, nearby structures should be spared. Function, growth, and nearby structures should be spared and preserved.[10] In this case, the lesion was excised surgically by excisional biopsy.

CONCLUSION

This case illustrates an atypical presentation of JOF with subtle findings on conventional imaging, contributing to diagnostic difficulty due to its underreported nature. It also emphasizes the superior role of CT in accurately delineating lesion characteristics. Early diagnosis, appropriate imaging, and complete surgical excision are essential to achieve optimal outcomes. This case adds to the limited literature on unusual presentations of JOF and underscores the need to consider JOF in atypical extraosseous-appearing mandibular lesions.

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