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Case Report Open Access

A Rare Case of Psammomatoid (Juvenile) Ossifying Fibroma of the Maxillary Sinus

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Abstract

A case of POF in the maxilla of a 10-year-old child is presented. This case is reported in view of its absence in the maxilla and the difficulty in diagnosis due to its radiological resemblance to lymphoma and osteosarcoma, further aggravated by the clinical scenario.

Keywords: Fibroosseous Lesion,; Psammomatoid Ossifying Fibroma; Tumor

Introduction

Ossifying fibroma is a rare fibro-osseous lesion that most commonly occurs in the craniofacial skeleton, with particular predilection for the nasal cavity, paranasal sinuses, and the orbit (Chung EM Radiographics 2008). Psammomatoid ossifying fibroma (POF), also known as juvenile (active) ossifying fibroma, is a variant of ossifying fibroma containing numerous calcified "psammomatoid" ossicles that histologically resemble psammoma bodies. A more common and related variant is the cementifying or cementoossifying fibroma, which occurs in gnathic bones (Slootweg PJ J Oral Pathol Med 1994, Kuta JA AJNR 1995, Kasliwal MK J Neurosurg Pediatrics 2011). Very few cases of POF have been described in the literature; most of them involve the paranasal sinuses or the orbit (Margo CE Ophtalmology 1985, Han MH AJNR 1991, Wenig BM Cancer 1995). We describe an unusual case of POF involving the maxillary sinus in a child who presented with swelling in the left facial region. The appearance of the lesion and its location presented a diagnostic challenge. Best of our knowledge, a POF involving the maxillary sinus with intracranial extension was not previously reported.

Case Report

History and examination

A 10-year-old male child presented to the Unit with a painless, progressive swelling of the left face for 3 months following the extraction of first primary molar tooth. Extraoral examination demonstrated a moderately large left facial mass. There was no facial anesthesia. Extraocular eye muscle movements were normal. Intraoral examination revealed gross expansion of the left maxillary alveolar process that is extending to the hard palate. There was minimal tenderness on palpation. The left maxillary molar and premolar teeth were loose. There were no palpable cervical or submandibular lymph nodes. His medical history was insignificant. He had no other complaints.

Imaging

Panoramic radiograph showed a homogeneously hyperdense whole left maxilla and maxillary sinus, and bone destruction and invasion of the molar part of the left maxilla, zygomatic buttress and part of the zygomatic bone. The left maxillary third molar and second premolar teeth were drifting. Left primary second molars showed signs of inflammatory external root resorption (Figure 1). Facial MR Scan revealed a mixture of radiolucency and radiodensity, and a well-defined osteolytic mass involving the left maxilla, maxillary sinus, alveolar bone and nasal cavity. There was destruction of the medial and antero-lateral walls, and the roof of the left maxillary sinus. The inferior orbital bone and cranial base were partially involved (Figure 2A and B). The chest radiograph was normal, also. The differential diagnosis included the following: metastases, lymphoma, osteosarcoma, cementoossifying fibroma, and trabecular ossifying fibroma.

Biopsy findings

The child underwent insicional biopsy to clarify the diagnosis. Histological studies revealed the presence of a POF characterized by a benign fibrous proliferation with admixed calcified psammomatoid ossicles (Figure 3). Although the lesion was benign, a complete excision

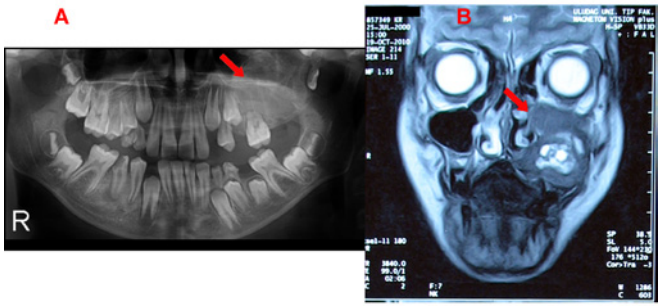

Figure 1: Panoramic view of the presented JOF lesion (indicated by a solid arrow).

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and osseous reconstruction were planned to prevent further expansion and to restore her normal facial contour.

Operation

The tumor was exposed through and a complete excision was performed through a transoral approach. A left total maxillectomy was made. Additionally, the orbital floor invaded by the lesion was also removed. The defect of the orbital floor was bridged using a prolene mesh to reconstruct the facial contour and to support the orbital contents. The mesh was fixed to the residual zygoma laterally, and to the nasal bone and edge of the anterior nasal aperture medially. The defect which was occurred following maxillectomy was covered with a split-thickness graft.

Postoperative course

The patient had an excellent outcome, with very good cosmetic reconstruction of the facial (orbital) defect. Following the operation an obstructer appliance was fabricated and placed in to acquire proper oral function and speech and to seal the oral cavity from the nasal space, (Figure 4 A-C). Patient has been scheduled for 6-months clinical and imaging follow-ups to prevent further recurrence of the tumor.

Discussion

Review of the literature shows that it is difficult to establish definitive diagnosis criteria for POF. The main characteristics are: a patient between 5-15 years of age, the location of the tumor, the radiologic pattern [2,7]. It comprises 2 % of oral tumors in children (Sun G, Chen X, 2007). Recurrence has been reported in as many as 28 % of patients with mandibular ossifying fibromas. However, the recurrence rate of the lesion in the maxilla is unknown, but it is likely to be higher because of the greater difficulty of its surgical removal and larger size at the time of presentation. This lesion is often difficult to diagnose, clinically, radiologically and histopathologically, also.

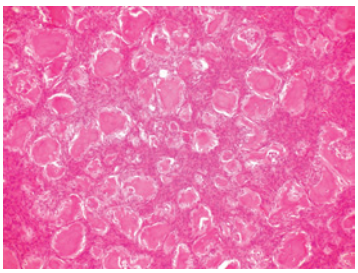

Figure 2: Facial MR Scan the presented JOF lesion (indicated by a solid arrow)..

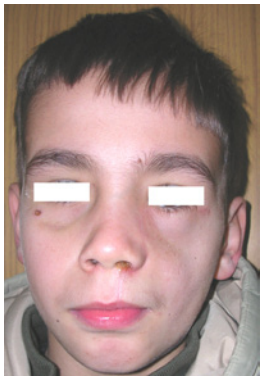

Figure 3: Postoperative facial apperance.

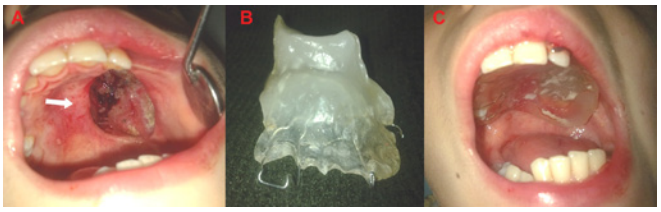

Figure 4: Postoperative intraoral view and appliance placed in.

Clinically, this lesion has in general a more aggressive growth rate than ossifying fibroma [5]. It is usually asymptomatic, and no significant sex predilection has been reported in the literature [5]. Rare cases of POF have been reported in paranasal sinuses, orbit, and fronthoethmoid bones (Chung EM Radiographics 2008). Its common variant cementoossifying fibroma which may develop in jaws was also reported in the literature (Kuta JA AJNR 1995). However, this reported case was the first POF case from which originated maxillary sinus in the literature as far as the authors have known. Generally, I the first sign of the tumor was expected to be swelling of the jaw bone as was in the present case. When the orbital bone and paranasal sinuses are involved, the patients may develop exophthalmos, bulbar displacement and nasal obstruction. However, the patient had no other complaints but swelling in this case.

The tumor has an extremely variable radiographic appearance, depending on its stage of development (Sarwar HG, Jindal MK 2008; Thankappan S, Nair S 2009). On plain radiograph such as panoramic radiograph or Water's view, it can appear radiolucent, radiopaque, or of mixed opacity depending on the degree of calcification of the lesion (Rinaggio J, Land M 2003). The MR appearance of these lesions can vary depending on the internal architecture, with enhancement of the solid portions of the lesion (Wenig BM Radiol Clin North Am 1998). Our patient had further diagnostic considerations due to the absence of involvement of the maxilla by POF.

Histologically, the lamellae are of variable shape, from trabecular to more osteoid. Hemorrhage is not prominent and when present is scattered rather than seen as foci, the latter being common in central giant cell granuloma. No obvious islands of epithelium or individual epithelial cells are present [9] as seen in odontogenic fibroma. The stroma shows focal areas with stromal cysts and occasional mitoses. Cementicle-like particles are not seen. At the periphery of the lesion, cortical and reactive bone is sometimes seen. Therefore, differential diagnosis includes central osteoma, ameloblastic fibroma, ameloblastoma, odontogenic cyst, vascular lesion, and giant cell lesions with a mandatory pathologic study because it is largely based on the nature of the calcified products of the tumor [5].

The clinical management and prognosis of POF are unclear. The recurrence rate ranges from 30 % to 58 % [3,4]. Conservative surgery or radical en-bloc resection are suggested. A conservative surgical excision (enucleation or curettage) also has been described as the treatment of choice for small lesions of POF [5] Rinaggio J, Land

M 2003). However, immediate recurrence characterized by a high aggressive growth rate and the absence of a distinct separation between tumor and the adjacent bone requires extensive surgery, with wide demolition of the involved bone [8,10]. In the present case, because the lesion had grown to a massive size and caused cosmetic and functional problems, a left total maxillectomy was performed. A prolene mesh was used to reconstruct the facial contour and achieve a satisfactory facial appearance. Following one month after surgery obstruction appliance was fabricated to close the palatal defect and to acquire acceptable oral function.

POF remains a rare diagnosis in expansile bony lesions of the cranium. POF involving maxilla had not been reported previously. Awareness of its cranial presentation and radiological and pathological findings may facilitate diagnosis and appropriate treatment. Good surgical reconstruction and orodental rehabilitation with teamwork are equally important.

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