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Maxillary psammomatoid juvenile ossifying fibroma: A rare clinicopathological entity

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Abstract

The juvenile psammomatoid ossifying fibroma (JPOF) is a rare benign fibroosseous neoplasm of the craniofacial bones. It has predilection for sinusal locations, in young patients, with aggressive behavior and high recurrence rate. Surgical intervention is needed to secure a complete resection of the neoplasm. We present one unusual case of a young man presenting with a manifestation of JPOF in the maxilla, an unusual location, that was approached and completely removed via Caldwell-Luc approach. Early detection avoided exaggerated distortion of the anatomy, allowing an excision without complex reconstruction needs, and no recurrences in the postoperative follow-up.

Keywords: Juvenile ossifying fibroma; Craniofacial; Caldwell-Luc; Case report.

Introduction

Juvenile psammomatoid ossifying fibroma (JPOF) is a rare benign, although aggressive, fibro-osseous neoplasm of the craniofacial bones. It is a subtype of juvenile ossifying fibroma with particular characteristics, since it is most commonly seen in early ages, with most cases reported in young teenage years, and has no gender predominance. The most common location is the paranasal sinus [1] with less common locations identified in the jaws (mandible and maxilla) and temporal bone, with male predominance in these locations and a more aggressive behaviour [2,3]. The predilection for certain locations distinguishes from the trabecular type that predominantly affects the jaws [4]. On CT the findings are consistent, a well delimitated mass with a thick wall of bone density, but variable density and multiloculated appearance inside and calcified areas. These imagiological findings are important in the jaws, to differentiate from fibrous dysplasia – without well-demarcated borders –, that has a limited clinical course [3]. Surgical intervention is needed to secure a complete resection of the neoplasm, with preservation of as much anatomical structures as possible, especially in younger ages when bone growth is a concern. The aggressive growth and tendency to recur is age related, and long follow-up periods must be considered, although there is no standard protocol [5]. The aim of this article is to report a clinical case of one unusual case of JPOF in the maxilla.

Case presentation

We report a case of a sixteen-year-old boy presenting to the emergency room with a painless swelling in the left maxillary area just near the pyramidal aperture, with continuous growth reported in the previous three months. No relevant family and medical history. There was no history of toothache, tooth treatment or discharge of purulent content. At clinical exam there was a mild but visible distortion of the left nasolabial fold and at examination of the oral cavity a hard palate cortical expansion was identifiable (Figure 1).

A facial CT (Figure 2) scan was requested, showing an apparently round cystic lesion, well defined, with 3.3 x 3 cm, in the second quadrant motivating a gap between 2.4 and 2.5 teeth, remodeling the floor of the nasal cavity was found as well as **Citation:** Eiró V, Ferreira M, Mendes C, Gigante T, Antunes SM et.al. Maxillary psammomatoid juvenile ossifying fibroma: A rare clinicopathological entity. J Clin Images Med Case Rep. 2024; 5(6): 3135.



Figure 1: Ipsilateral hard palate expansion.



Figure 4: Intraoperative photograph of the specimen.

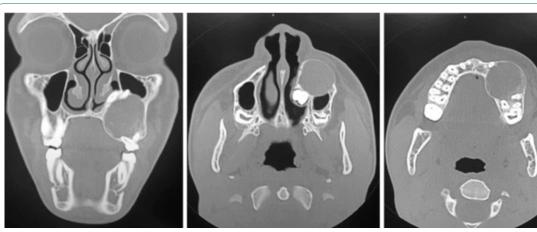


Figure 2: CT scans-second quadrant mass with remodeling the floor of the nasal cavity and obliteration of the maxillary sinus. An impacted tooth is also identifiable.



Figure 3: Intraoperative photographs of complete removal by Caldwell-Luc approach.

obliteration of the maxillary sinus. A few calcifications were noticeable inside of the mass. The diagnostic hypothesis of odontogenic keratocystic tumor, myxoma and ameloblastoma were also contemplated.

Considering its volume and continued rapid growth, the surgical excision was programmed. In the operating room, under general anesthesia, a vestibular approach was performed with external approach (Caldwell-Luc technique) and removal of what proved to be a solid, rich in connective tissue, mass. Curettage, and removal of an impacted tooth in the sinus were also executed (Figure 3). During the intervention, the cortical conti-

nuity was preserved, and no bone reconstruction was needed. The surgery and postoperative period were uneventful, and the patient was discharged on the second postoperative day.

There were no complications during the recovery period. The specimen (Figure 4) was sent for histopathologic analysis and the concluded diagnostic was juvenile psammomatoid ossifying fibroma. At nine months of follow up, there were no recurrences or new lesions.

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Conclusions

Although it is a benign pathology with no description of malignant transformation in literature, its aggressiveness and expansile behavior can interfere with function (obstructive airway, visual changes), bacterial infections (recurrent sinusitis or associated with intracranial extension [1]) and psychosocial interactions (visible deformity). Therefore, the treatment of choice is early surgical excision, before the remodeling of anatomic structures nearby dictates a need for a more complex reconstruction, with higher morbidity associated. Special care must be taken to completely remove the entire lesion, to avoid recurrences. If so, it is associated with very good prognosis. Notes The authors have declared that no competing interests exist.

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