Autopsy Case Reports

Article / Clinical Case Report

Odontogenic myxoma in a child treated with enucleation and curettage

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ABSTRACT

Odontogenic myxoma is an aggressive benign odontogenic tumor, accounting for 3-6% of all the odontogenic tumors in adults. The incidence among children is lower. Due to its clinical behavior, there is no consensus on the best treatment. In this paper, the authors report the case of a 9-year-old girl with the diagnosis of odontogenic myxoma. The panoramic X-ray showed an extensive radiolucent lesion involving the left mandibular body causing teeth displacement. The treatment consisted of tumor enucleation followed by vigorous curettage of the bone walls. Both the base of the mandible and the inferior alveolar nerve were preserved. The patient is asymptomatic after 6 months of surgery. The age of the patient and the radiographic features were taken into account when deciding in favor of the conservative treatment.

Keywords
Myxoma; Odontogenic Tumors; Pediatric Dentistry; Diagnosis, Oral.

INTRODUCTION

Odontogenic myxoma (OM) is an uncommon benign odontogenic tumor of mesenchymal origin, which affects the maxillary bones without gender predilection. OM is mostly incident in the third decade of life, accounting for about 3-6% of all the odontogenic tumors in adults and is even rarer in children. It is a locally aggressive tumor with a recurrence rate of 25%. Radiographically, OM presents as a well-delimited unilocular or multilocular osteolytic lesion, in which the appearance of "soap bubble" or "honeycomb" can be observed. Expansion of the cortical bone and teeth displacement are also common findings. The ameloblastoma is the main clinical-radiographic differential diagnosis.[1-3]

The surgical approach to OM treatment currently ranges from conservative tumor enucleation to aggressive resections such as segmental mandibulectomy or maxillectomy. The best therapeutic option for pediatric patients is another matter of debate in the literature.[3,4]

The objective of the present study was to report a case of OM in a child and discuss the advantages of the treatment modalities.

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CASE REPORT

A 9-year-old girl attended our clinic complaining of painless growth of her mandible over the previous year, which caused mild facial asymmetry. On intraoral examination, the vestibular bone of the left mandibular body was expanded. The panoramic X-ray showed an extensive radiolucent lesion involving the left mandibular body. Not only did the tumor cause teeth displacement, but some thin bony septa were also observed within the lesion (Figure 1).

The computed tomography images showed a hypodense and well-delimited lesion in the left mandible. Large expansion of the buccal and lingual plates was observed, but no perforation of the cortical bones was present (Figure 2).

The diagnostic hypotheses were ameloblastic fibroma, OM, and ameloblastoma. An incisional biopsy was performed, and a yellowish gelatinous tissue was obtained for laboratory analysis (Figure 3).

The histopathological aspect showed a myxomatous tissue with fusiform cells interspersed by thin collagen fibers (Figure 4). According to clinical, imaging and histopathological features, the final diagnosis was OM.

Before the surgery, endodontic treatment of the left lateral incisor and first premolar was performed because of the periapical involvement of both teeth. The surgical treatment consisted of total enucleation and vigorous curettage of the bone walls. The inferior alveolar nerve and the lower mandibular cortex were preserved. The healing of the surgical wound occurred by secondary intention due to dehiscence. However, no signs of infection were observed. The panoramic radiograph on the 4th month of follow-up showed adequate bone formation without signs of recurrence (Figure 5). The patient is asymptomatic after 6 months of treatment.

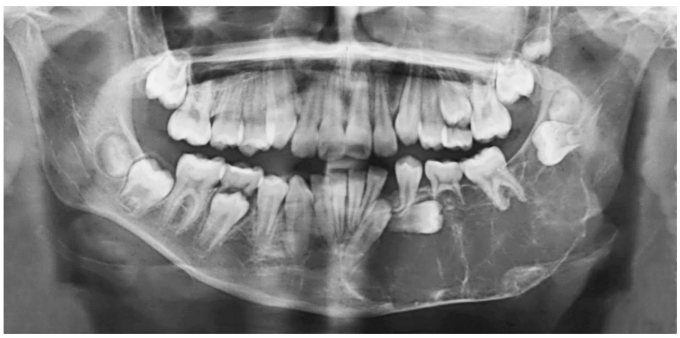

Figure 1. Panoramic radiograph showing an extensive unilocular radiolucent lesion in the left mandibular body exhibiting thin bone septa with "honeycomb" aspect and tooth displacements.

DISCUSSION

OM arises from the odontogenic mesenchymal tissue, histologically resembling the follicle and dental papilla. It is characterized by stellate and

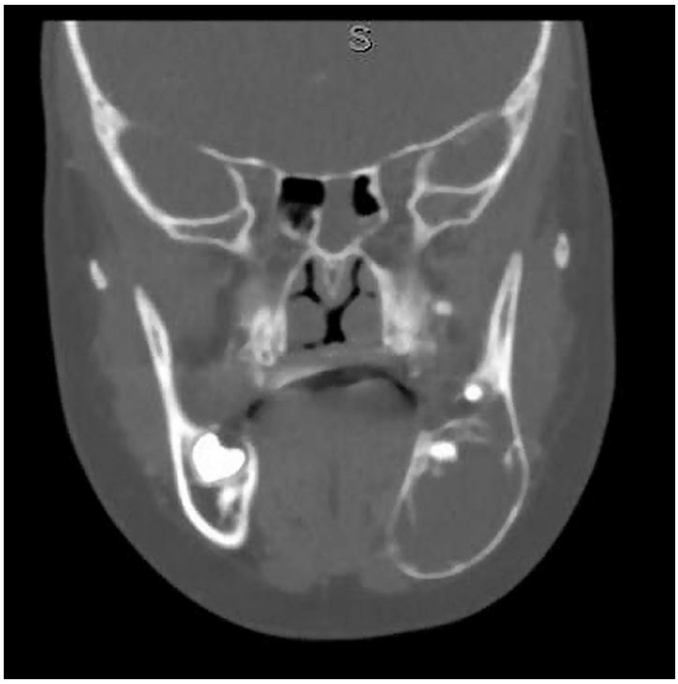

Figure 2. Computed tomography (coronal section - bone window) presenting a hypodense lesion in the left mandibular body, which expands the cortical bones without perforation.

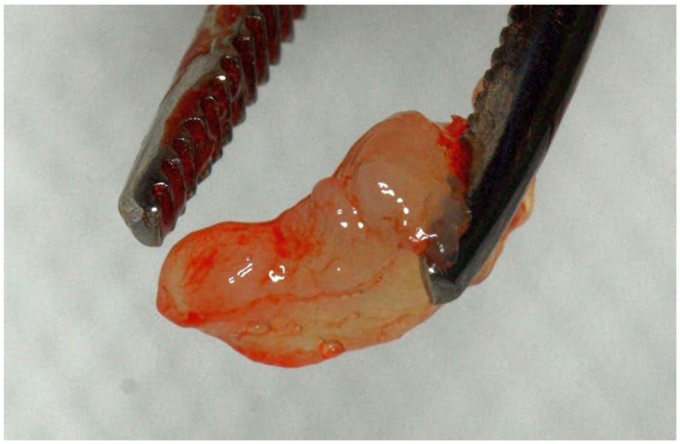

Figure 3. Gross examination of the biopsy specimen showing a gelatinous and yellowish tissue (measuring 0.5 cm in its longest axis).

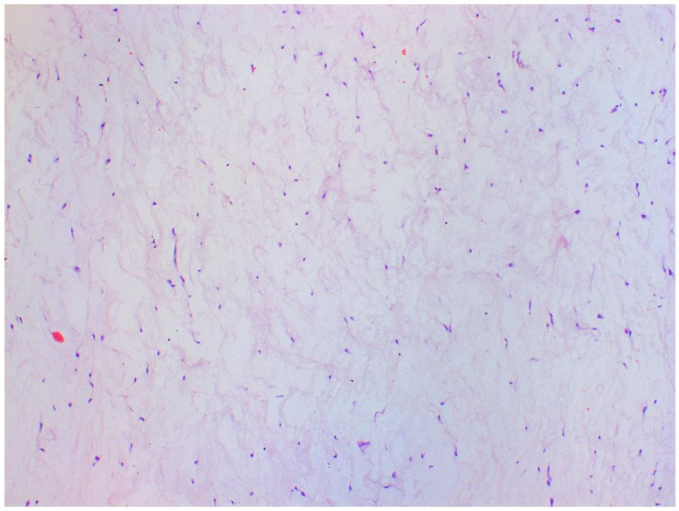

Figure 4. The myxomatous tissue with fusiform cells interspersed by thin collagen fibers (H&E, 100X).

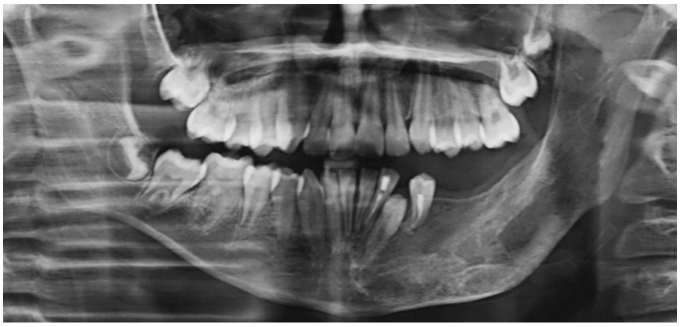

Figure 5. Panoramic radiograph after 4 months of the treatment. Bone formation can be observed in the posterior region of the mandible.

spindle-shaped cells dispersed in an abundant myxoid extracellular matrix. Clinically, it is locally aggressive and infiltrates the adjacent structures. The maxillary lesions usually extend to the maxillary sinus, and the mandibular ones surround the mandibular canal. Such a tumor is sporadic in children under 12 years old, being more common between 20 and 40 years of age. Radiographically, OM presents as a well-delimited osteolytic lesion. The "soap-bubble, honeycomb or tennis-racket trabeculation" appearance can be observed.[1-3,5-8] The case presented herein shows an OM affecting the mandible of a 9yo girl. The tumor caused a mild facial asymmetry on the left side of the face due to the enlargement of the left mandibular body, but there was no alteration of the oral mucosa.

Due to the OM biological behavior, some benign or even malignant odontogenic tumors and other maxillary lesions such as central giant cell granuloma are included as differential diagnoses.[3] In the present case, ameloblastic fibroma (AF) was one of the hypotheses due to the patient's age. Such a tumor is more frequent in young patients, presents slow and painless growth, and the posterior region of the mandible is mostly affected. The AF radiographic images vary from well-defined and small unilocular radiolucent lesions to a multilocular pattern. Impacted teeth (usually the first or the second permanent molar) are present in 80% of cases, and root resorption and cortical perforation are uncommon.[5] Ameloblastoma (AB) was another highly-considered hypothesis, which occurs more frequently in the posterior region of the mandible, and the "soap bubble" and "honeycomb" radiographic patterns are also usually present. However, the age of our patient was out of the ameloblastoma epidemiologic features. Moreover, root reabsorption, which is common in large ameloblastoma, was not observed in our case.[5,9] During the incisional biopsy, a bright-yellowish and gelatinous tissue was removed, which indicates a striking macroscopic feature of OM (Figure 2). This finding associated with radiographic features led us to consider OM as the main diagnostic hypothesis.

The therapeutic option for odontogenic tumors in children should consider aesthetic and functional aspects, especially for OM, which is a locally aggressive benign tumor. The surgical approach ranges from conservative enucleation to radical resection with adequate surgical margins to avoid recurrences.[4,7] Kansy et al.[3] performed a literature review of OM and added 4 cases affecting the maxilla. These authors showed a trend to perform radical surgeries in adults, while conservative approaches such as enucleation and curettage of the lesion are recommended for children. The recurrence rates were similar comparing enucleation and partial maxillectomy.[3] Subramaniam et al.[4] warned surgeons of the impact on both the growth and the function of the jaw bones and surrounding structures when radical procedures are performed in children.

Also, conservative management may result in acceptable recurrence rates with less morbidity. In this case, however, a long-term close follow-up is mandatory, as the presence of cortical (lingual and basal) remnants allowed the conservative approach represented by enucleation and vigorous curettage. After 6 months, a satisfactory bone formation was

observed, including the height of the mandible bone (Figure 5). The patient is currently under a close follow-up.

In conclusion, OM exhibits aggressive local behavior, and large resections are more frequently indicated. However, in exceptional cases such as the subject of this case report, a conservative approach (enucleation and vigorous curettage) can be an option to avoid facial and functional deformities. A close follow-up is mandatory to diagnose possible recurrences.

The patient's father signed an informed consent authorizing the publication of this report as well as the images. The present case report is exempt from approval from an ethics committee.

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Author contributions: Oliveira SV, Alves FA and Gallo CB were in charge of the patient's care and wrote the manuscript. Rocha AC and Ceccheti MM. performed the surgical procedure and follow-up the patient. All authors collectively proofread and approved the manuscript for publication.

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