



Odontogenic Myxoma in a Pediatric Patient: A Case Report

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Abstract

Odontogenic myxoma (OM) is a benign intraosseous tumor, but locally invasive, having a high rate of recurrence. It preferably locates in posterior mandibular regions, followed by the incisors, maxillary and; exceptionally, the mandibular condyle. Odontogenic myxoma can be found most often in young adults aged 25 - 35 years although lesions may occur over a lifetime, reporting the lowest prevalence in childhood with a predilection for permanent dentition. This paper presents a case of a three-year-old boy who visits the dentist because some mandibular teeth have not erupted. Panoramic radiograph and CBCT exams were performed. The tumor was radiological and histologically diagnosed as odontogenic myxoma. It was decided to do conservative surgery and it has been controlled for 4 years with favorable results and without imaging signs of relapse. This case demonstrates the unpredictability of a pathological maxillo-facial lesion, and leads to suggesting early control of the dental development process in pediatric patients.

Subject Areas

Dentistry

Keywords

Odontogenic Myxoma, Odontogenic Tumors, Pediatric Patient, Cone-Beam Computed Tomography, Case Report

1. Introduction

Odontogenic myxoma (OM) is a benign intraosseous neoplasia, not very frequent, [1] but with high infiltrative potential that makes it a locally invasive tumor. It belongs to

the Classification of Odontogenic Tumors, to the subgroup derived from ectomesenchyme [2] [3] [4] [5].

It appears as a slow-growing lesion, but with infiltrative potential that can produce cortical bone expansion, causing large bone destruction at its later stages. Moreover, it shows recurrence rates from about 5% to 10% although there are no currently reliable data in the literature [4] [6].

OM is an uncommon disease; some studies reveal that it represents between 3.3% - 25.7% of all odontogenic tumors in adults, and between 8.5% - 11.6% in children [4] [7] [5] [6] [8]. In Chile, its prevalence is considered to be 8.8% among all odontogenic tumors at all age ranges [9].

It has been reported in several studies that its prevalence in children is very low. Fang, Shi and Sun, in 2014, reported that in pediatric patients, OM barely reaches 3.6% of the cases, being most common over the permanent dentition period [10]. The lesions in the maxilla tend to obliterate the maxillary sinuses as an early feature [2] [3] [4].

Smaller lesions may be asymptomatic, and they are only found through radiographic examination [2] [3]. Larger lesions are often associated with painless, slow-growing bone expansion, which can cause perforation of cortical bone, resulting in swelling and facial deformity. Moreover, we may find displacement of teeth and even root resorption, and these lesions may be associated with retained or missing teeth [4] [7] [11].

Radiographically, the lesion can show different appearances, which can be unilocular or multilocular. A radiolucent lesion with edges that can be defined or diffused, is observed. Sometimes, they may have a radiopaque appearance, particularly, lesions associated with the maxillary sinus [1]-[14].

It is important to determine the exact diagnosis to be able to define the prognosis and treatment plan to perform. The aim of this paper is to emphasize the importance of making timely imaging controls in pediatric patients given the unpredictable nature of many pathological lesions of the jaws.

2. Case Report

Three-year-old boy patient visits the dentist because his deciduous mandibular left lateral incisor has not erupted.

Clinical examination shows absence of the tooth, normal lining mucosa, and increased volume in buccal and lingual bone plates, the lingual one being more compromised. He is referred to an initial radiological examination, requesting a Panoramic radiograph (Figure 1).

The Panoramic radiograph is taken on October 11, 2012, showing a multilocular radiolucent lesion that goes from distal of tooth bud 31 to tooth bud 34 zone. It produces caudal displacement of tooth bud 32, and distal displacement of tooth bud 33, plus embedded tooth 72. In cranio-caudal direction, it compromises alveolar bone ridges until the inferior border of the mandible (Figure 1).

Additionally, a Cone-Beam volumetric tomography (CBCT) is performed, in order to evaluate local and general compromise of the lesion (Figure 2 and Figure 3).



Figure 1. Panoramic radiograph. First exam taken to the patient where the finding of the lesion is made. It shows a multilocular radiolucent lesion that produces displacement of tooth buds and compromises alveolar bone ridges until the inferior border of the mandible.



Figure 2. Cone beam CT exam, panoramic view.



Figure 3. Cone beam CT exam, sagittal view.

Imaging confirms the presence of a hypodense multilocular area involving tooth buds 31-32-33-34, and the diagnostic hypothesis proposed is odontogenic myxoma (**Figure 2** and **Figure 3**).

The histopathological study, in which part of the lesion and the impacted deciduous tooth are removed, is performed. The macroscopic sample is divided into two parts, which are sent to two histopathology laboratories. The first biopsy delivers its result on October 18, 2012, confirming the odontogenic myxoma diagnosis: *“Tumorous lesion composed of ovoid and spindled cells, merged into a myxoidstroma. Dilated and congested vessels, and some zones of odontogenic-like epithelial proliferation are seen. There is no significant cell atypia, necrosis, or mitotic activity. The sample includes a tooth with preserved aspect.”*

The second histopathological study of the original sample is delivered on November 16, 2012, confirming the previous radiographic and histological diagnosis, *“Spindled cellular tissue proliferation, with elongated stellate nuclei, with abundant ground substance, some bundles of collagen fibers, small capillaries; and which presented some bundles of S100 positive nerve fibers at the periphery, but the cellular proliferation was mostly negative”*.

It was not possible to obtain the histological images of this particular clinical case, but a characteristic reference cut of this lesion is shown [15] (**Figure 4**).

In the surgical procedure, curettage is performed conserving involved teeth except for one that was removed during the biopsy, and the progress of the lesion would be supervised.

Five months later, a follow-up with a Panoramic radiograph is performed, showing bone tissue neoformation (**Figure 5**).

The patient has been under clinical and imaging follow-up for 2 years (every 3 - 4 months) without any apparent changes that suggest lesion recurrence; neoformation and bone restructuring are seen. Two years after diagnosis, he is examined through CBCT, which confirms neoformation and bone restructuring (**Figure 6**).

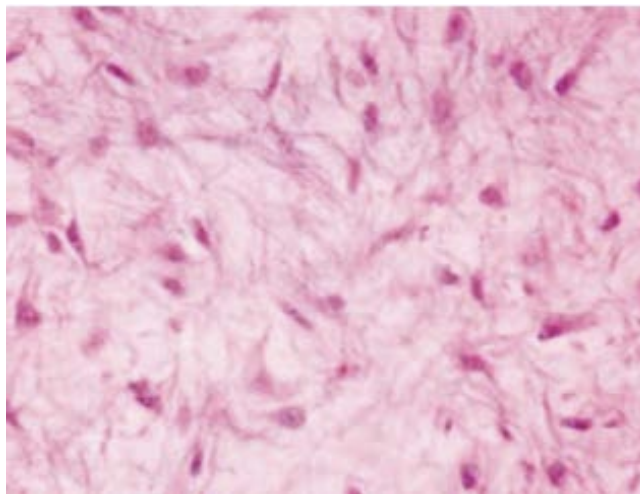


Figure 4. Histopathological image of odontogenic myxoma (referencial) [15].

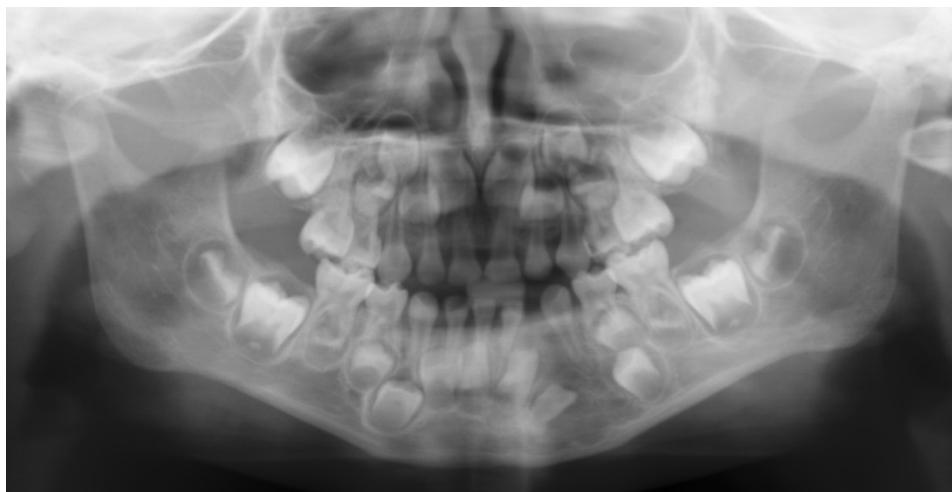


Figure 5. Panoramic radiograph taken 5 months after surgical procedure showing bone tissue neoformation.



Figure 6. Cone beam CT exam taken as follow-up 2 years after surgery.

Radiographic follow-up every 3 to 4 months through Panoramic radiograph is suggested to control potential recurrence.

Currently (2016), the patient is 7 years old, healthy, dynamic and not medically compromised. Panoramic radiograph shows the four year follow-up without signs of recurrence (**Figure 7**). He goes to regular dental check-ups.

3. Discussion

Literature review shows that odontogenic myxoma is an uncommon tumor [4] [8] [9] [10], found mainly in young adults, without many case reports in children [2] [4] [10] [12] [13]. According to the scientific evidence, OM mainly affects women although some authors report that there is no gender preference [2] [3] [5] [13]. OM can be found in all age groups, but with an incidence peak between the second and third decade (25 - 35 years) [2] [4] [8] [16].

In the literature, there are few case reports of pediatric patients, Schwenzler-Zim-



Figure 7. 2016 panoramic radiograph taken 4 years after surgical procedure.

merer K, *et al.* reported, in 2012, two cases of children between 11 and 12 months old [4]. OM can be found both in the maxilla and the mandible, but it preferably locates in the posterior region of the mandible, followed by the incisor region; then, in the maxilla and; exceptionally, in the mandibular condyle [7] [9].

This clinical case in a 3-year-old patient shows how unpredictable odontogenic lesions can be, in this respect. In most cases, the diagnosis is made at an older age when the lesion has already shown clinical features, that is, it has reached important growth [14]. In some instances, clinical growth can be rapid, which is probably related to myxoid substance accumulation in the tumor [2] [3]. When there is sinus obliteration, this lesion can be similar to a nasal polyposis, and may be confused with it [2].

The radiographic features of OM are hard to differentiate from other odontogenic and non-odontogenic tumors that can be found in the maxillae [11] [13] [14] [16]. However, its characteristic appearance formed by a multilocular radiolucent zone described as “soap bubbles” or “honeycomb”, can be observed [1]-[13] [16].

Computed tomography (CT) and magnetic resonance imaging (MRI) allow defining the extent of the lesion more precisely, and analyzing the different dimensions of its behavior (bone plate expansion, involvement of teeth, and compromise of neighboring structures, among others) [17].

As mentioned before, the image of the lesion, both in the radiograph and in the CT, is radiolucent, generally multilocular, but in the MRI there are a characteristic medium intensity signal on T1, and a hyperintense signal on T2 [17]. Both the radiograph and the CT or MRI should correlate with the only exam that can provide accuracy when diagnosing a bone lesion, that is, the histopathological study [3]-[14] [16] [17].

In this case, the tumor was found because the patient was referred to a routine radiographic exam to follow up the dental development process, which permitted prompt and less invasive surgical treatment, improving case prognosis as seen on the following check-ups [14].

4. Conclusion

Even though it is an uncommon tumor in children [3] [6] [7] [8] [16], odontogenic myxoma is a locally aggressive lesion, therefore the importance of periodic dental follow-up of the pediatric patient, especially if there are eruption anomalies, or any clinical, or radiographic findings that make us suspect.

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