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An expanded odontogenic myxoma in maxilla

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SUMMARY

Odontogenic myxomas are considered to be a benign odontogenic tumor with locally aggressive behavior, non-metastasizing neoplasm of the jaw bones. It derives from the dental mesenchyme or periodontal ligament. Despite the benign nature of these lesions, there is a high rate of local recurrence after curettage alone and in certain cases requires adequate resection. This paper describes a case of a large odontogenic myxoma in the maxilla, emphasizing a discussion on the differential diagnosis related to radiological findings and the surgical treatment.

Key words: odontogenic myxoma, odontogenic tumors, maxilla.

INTRODUCTION

Myxomas can be found in various sites in the body including the skin and subcutaneous tissue, heart (mainly in the left atrium), and also in various sites of the head and neck. Odontogenic myxoma (OM) of the jaw was first described by Thoma and Goldman [1] in 1947. In the international histological classification of odontogenic tumors, OM is defined as a benign odontogenic tumor of mesenchymal origin that is locally invasive and consists of rounded and angular cells that lie in abundant mucoid stroma [2]. It is a non-encapsulated benign tumor of the jaws that occurs very rarely. They can be divided into 2 groups: tumors that arise specifically in jawbones (the most common type) and those that arise in soft tissues of that area. OM is commonly described as a slow growing tumor and generally symptomless. The origin of OM is believed to be odontogenic ectomesenchyme of a developing tooth or undifferentiated mesenchymal cells in the periodontal ligament [3]. Some authors had previously associated its origin with a myxomatous change of an odontogenic fibroma [1,2] or residual foci of embryonic tissue.

In Africa OM is seen to be the second commonest odontogenic tumor after ameloblastoma occurring with relative frequencies between 1 and 19% [4-7]. In Asia, Europe and America relative frequencies between 0.5 and 17.7% have been reported [3,8-13].

OM is most commonly seen in patients who are above the age of 10 and below 40 years [14-17], but an OM can occur at any age from childhood to the elderly [17]. In general, OM is rare in children below the age of 10 and in adults above the age 50 [3,16]. However, Kaffe et al. [18] in a review of 164 cases of odontogenic myxomas noted that approximately 7% occurred in the first decade of life. Similarly, Keszler et al. [3] in reviewing 367 cases of jaw myxomas reported an 8.4% occurrence in children less than 16 years of age. There are reports of OMs in patients under 10 years of age [3,19-21], with the youngest recorded case in a baby of 3 months [16]. Thus, one should consider odontogenic myxomas to be of higher frequency than once thought in the pediatric population [21]. Correlation between tumor size and age of patients was significant in the study of Noffke et al. [17]. Their incidence is similar in both sexes [3,22].

Although its most frequent location is the posterior mandible, other locations such as the incisive sector, upper maxilla and mandibular condyle must be considered [17]. The premolar–first molar region is the site of predilection in the maxilla [17]. Odontogenic myxoma rarely crosses the midline. According to Simon et al. [16] the lesions occupy one side only (left or right), but Noffke et al. [17] found six mandibular lesions that crossed the midline, being probably size related. There are cases in the literature of different lesions in the mandible; one in the left and other in the right side [16].

In many cases, these lesions are diagnosed accidentally by a routine dental checkup [15]. However, as the lesion advances in size OM may present with

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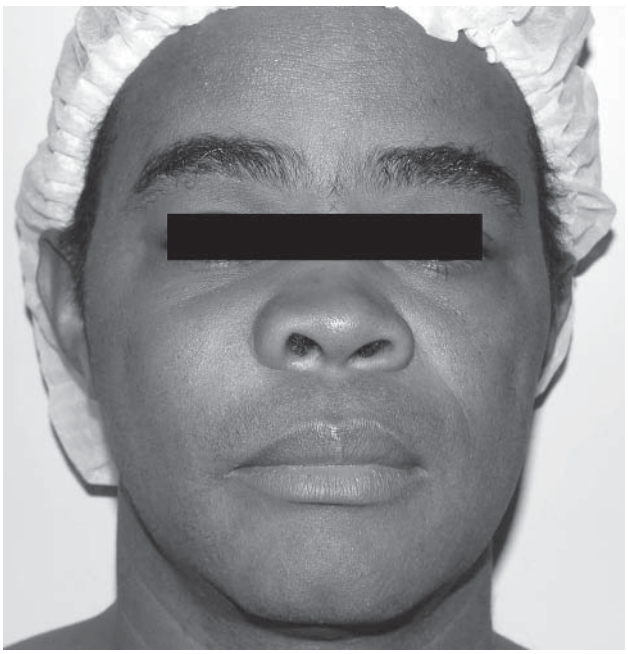

Fig. 1. Frontal view shows obliteration of the right naso-maxillary groove

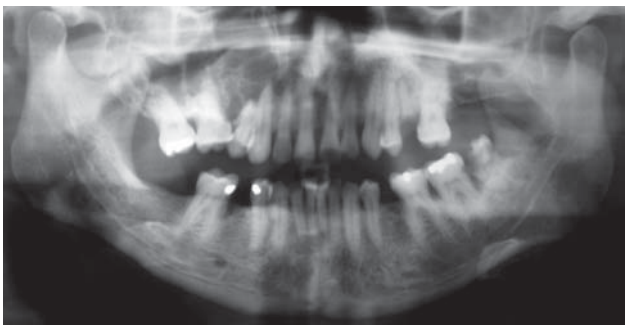

Fig. 2. Initial panoramic radiograph, showing multilocular lesion in the right maxilla and root resorption

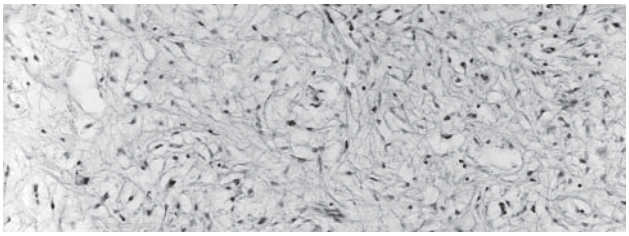

Fig. 3. Loosely arranged stellate, spindle-shaped and round cells and small islands of inactive odontogenic epithelial rests was scattered throughout the myxoid ground substance (HE, ×200)

disturbing symptoms. Bony perforation, with subsequent invasion into the soft tissues, pain, paresthesia, mobility of teeth and ulceration were among the other complaints that patients present with [4,16,23]. It was reported that involved teeth could be mobile but are usually vital [23].

OM is a slow growing tumor consisting of an accumulation of mucoid ground substance with little collagen, the amount of which determines whether it can be called a myxofibroma (also called fibromyxoma in the literature). The neoplasm grows because of this myxoid material [8]. Only occasionally does odontogenic myxoma show the presence of odontogenic epithelium or dystrophic calcification [2,3]. The tumors re usually interspersed with a variable number of tiny capillaries [16]. Atypical nuclei are often encountered, but cellular pleomorphism, prominent nucleoli and mitotic figures are not generally seen. Apparently its clinical and radiological limits may not histologically represent the true limits of the tumor [16]. Immunohistochemically, it has been reported the presence of mesenchymal markers such as muscle-specific actine, vimentine and less frequently S-100 protein [15].

Radiologically OM may appear as a unilocular or multilocular radiolucency with a "honeycomb", "soap-bubble" or "tennis racket" pattern with cortical expansion and tooth displacement [3,15,18,23,24]. Lesions greater than 4 cm tend to be multiloculated, and smaller lesions tend to be uniloculated [18]. The radiolucency may have clearly defined borders or poorly defined diffuse borders [17,18]. Kaffe et al. [18] found radiological evidence of calcifications in 12.5% of 164 OMs published up to 1997. According to Noffke et al. [17] variations in radiographic presentation make a radiological differential interpretation of OM challenging because the radiographic features overlap with those of other benign and malignant neoplasms.

Some OMs may show a mixed radiopaque-radiolucent appearance [16,18], which was ascribed to the presence of foci of calcification [18,22]. It was suggested that this appearance may be due to residual bone and not to new bone formation, and therefore it was proposed that OM should be considered in the differential diagnosis of mixed radiolucent-radiopaque lesions [18]. A number of lesions should be included in the differential diagnoses including ameloblastoma, intraosseous haemangioma, aneurysmal bone cyst, glandular odontogenic cyst, central giant cell granuloma, cherubism, metastatic tumor and, in cases of unilocular lesions, simple cysts, and the odontogenic keratocyst, especially the multilocular type [17,24,25]. In older patients, the possibility of a malignancy should not be ruled out because radiographic features resembling osteosarcoma have been reported [24,26]. A biopsy is, therefore, necessary to ascertain an accurate diagnosis.

Despite the fact that odontogenic myxoma shows aggressive local growth it is believed that it never undergoes malignant transformation or gives rise to metastasis [22], but there are reports of presumed benign lesions that then follow an aggressive course with local recurrences and distant metastases [27]. Despite the benign nature of these lesions, there is a high rate of local recurrence after curettage alone and in certain cases requires adequate resection [28].

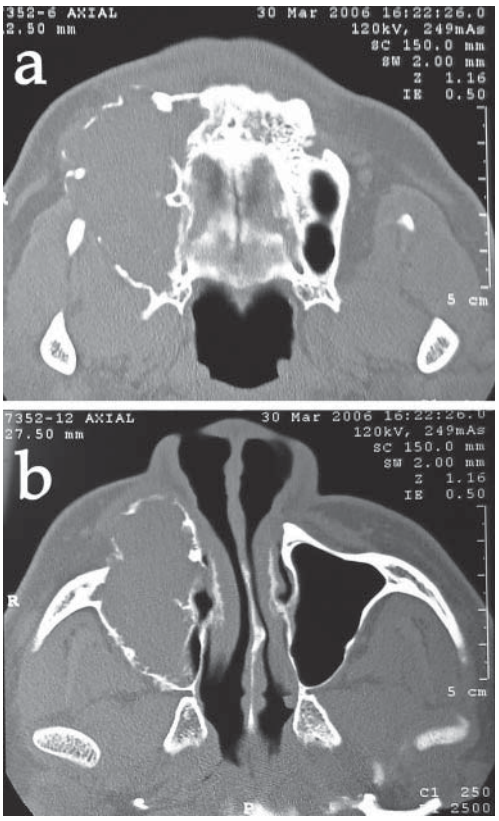

Fig. 4. Axial CT. Large mass involving the maxillary sinus, with destruction of the bony margins of the maxillary sinus (a). Little expansion to the nasal cavity (b).

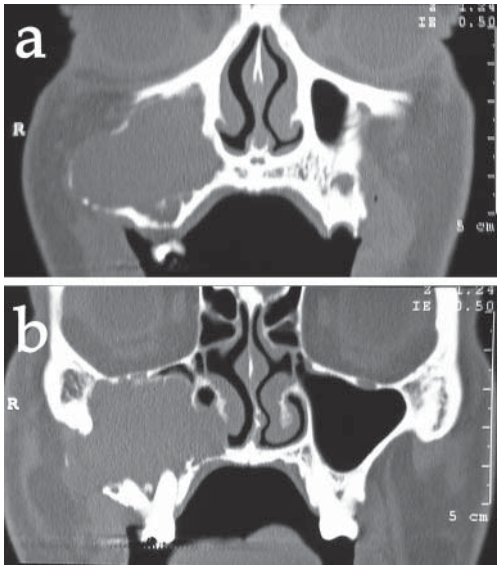

Fig. 5. Coronal CT. Expansion to the lateral right maxillary sinus wall (a). Destruction of the lateral nasal wall, invading the inferior nasal meatus (b).

The purpose of the present article is to present one case that was treated in the Department of Oral and Maxillofacial Surgery, at the Pontifícia Universidade Católica de Minas Gerais, Belo Horizonte, Brazil, emphasizing a discussion on the differential diagnosis related to radiological findings and the surgical treatment.

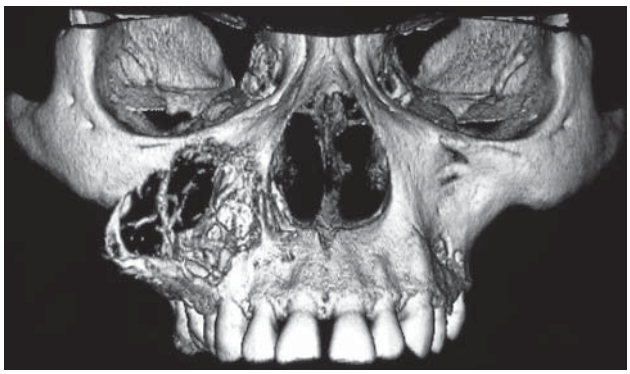

Fig. 6. Tridimensional CT

CASE REPORT

A 46-year old man was referred to our department with a history of a slowly enlarging right maxillary mass of 8-years duration. Physical examination revealed a unilateral swelling of the right middle-face, which produced a facial asymmetry, with obliteration of the right nasomaxillary groove (Figure 1). The patient denied bleeding, mucosal ulceration or sensory loss of the upper lip, oral structures or teeth, but had a partial right nasal obstruction. It was painful to palpation, but with no pain in mastication. An intraoral exam revealed no expansion at right palate while the mucosa overlying the area of the lesion was the same color and texture as the surrounding mucosa. The rest of the clinical head and neck examination was unremarkable. He had no general history of interest.

Initially, a panoramic radiograph was performed, which showed radiolucent lesions in the right maxilla with interlaced bone trabeculae resulting in a multilocular soap-bubble or honeycomb appearance (Figure 2). It also showed root resorption of the first right superior pre-molar. The differential diagnosis included odontogenic myxoma and central giant cell lesion. To establish a definitive diagnosis, an incisional biopsy was performed with the patient under local anesthesia. Microscopic examination demonstrated abundant loose myxoid tissue containing few collagen fibrils. The tumor was composed of loosely arranged stellate, spindle-shaped and round cells and small islands of inactive odontogenic epithelial rests was scattered throughout the myxoid ground substance (Figure 3 – Hematoxylin-eosin stain; original magnification ×200). The specimen was diagnosed as an odontogenic myxoma.

A computed tomography (CT) scan without contrast material with axial and coronal sections and tridimensional reconstruction was carried out to show the true extent of the lesion. The axial section showed a large mass involving the maxillary sinus, with destruction of the bony margins of the anterior and lateral right maxillary sinus wall (Figure 4a), and with little expan-

sion to the nasal cavity and no expansion the infratemporal fossa (Figure 4b). The coronal section showed an expansion to the lateral right maxillary sinus wall, with destruction of the bony margins (Figure 5a), with involvement of teeth, but without displacement. It also showed destruction of the lateral nasal wall, invading the inferior nasal meatus (Figure 5b). The tridimensional CT showed the right expanded maxillary area, and the multilocular nature of the lesion (Figure 6).

Under general anesthesia a Weber-Fergusson incision was made to expose the whole aspect of the lesion (Figure 7). The periosteum was elevated and the thinned anterior maxillary wall was exposed. All teeth from right superior canine to right superior second molar were removed together with the lesion, in one piece, that measured 65×50×40 mm (Figure 8). Macroscopically, this tumor was a soft, glistening, gelatinous mass.

After complete removal of the mass, the cavity borders were carefully osteotomized with a large spherical drill, to minimize the recidive chance. It was placed a titanium mesh to avoid complete collapse of the right cheek (Figure 9). There were no surgical complications. The patient's postoperative course was uneventful, and he was discharged from the hospital 2 days after the surgery. The patient was doing well, with good vision, no systemic or ocular problems and no radiological signals of recurrence fifty months after the surgery. After all this time there was some scar retraction. There was also a little loss of anterior and lateral zygomatic projection and a little loss of support of the soft tissues of the right hemi-face, even with the presence of titanium mesh, resulting in an increase in the opening between the eyelids of the right eye in rest position (Figure 10). However, the patient did not feel aesthetically impaired and did not want any additional surgery.

The patient had postoperatively a sensory loss in the area of innervation of the right infraorbital nerve, situation which remains until today, due to the removal of the distal part of this nerve together with the tumor mass.

There was a change in the relationship between the pupils. Before surgery (Figure 11a), the right pupil was slightly cranial to the left pupil, probably due to the slight elevation of the right orbital floor (see Figure 5b). After surgery, the right pupil was slightly caudal to the left pupil, probably due to the removal of the tumor mass and tissues retraction. The patient is currently using a superior removable partial denture without complaints.

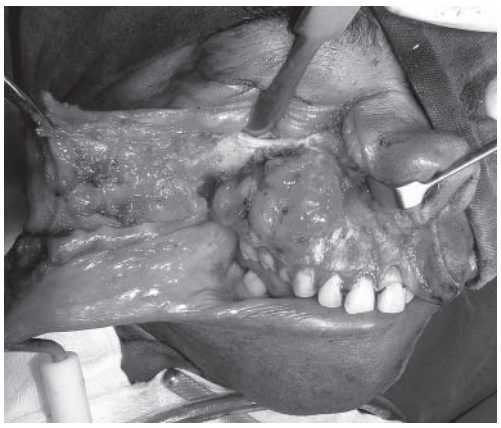

Fig. 7. Whole aspect of the lesion after a Weber-Fergusson incision

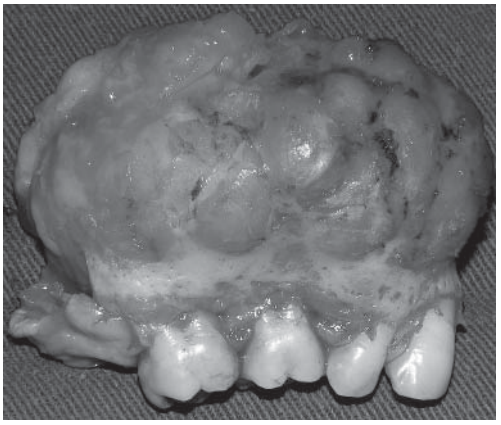

Fig. 8. Lesion removed in one piece

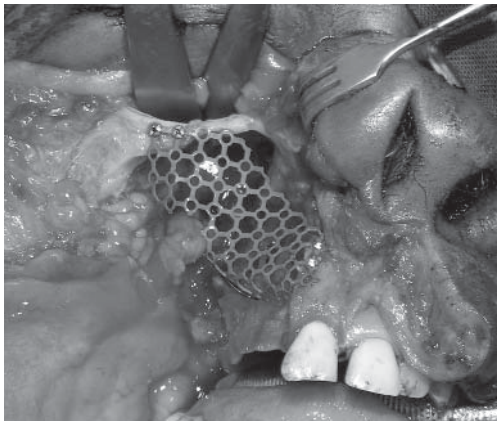

Fig. 9. Titanium mesh to avoid complete collapse of the right cheek.

DISCUSSION

OM is commonly described as a slow growing tumor and generally symptomless. There are also scattered reports of rapid enlargement [18,26]. In the present study was quite apparent that as it advances in size OM may present with disturbing symptoms. In general, the impression exists that the duration of symptoms is in accordance with the size of the tumour and magnitude of the symptoms [16]. Bony perforation, with subsequent invasion into the soft tissues, which is reported to be common in large OMs, was also observed in this patient. Paresthesia, mobility of teeth and ulceration were among the other complaints that patients usually presented with, but were not present in our case.

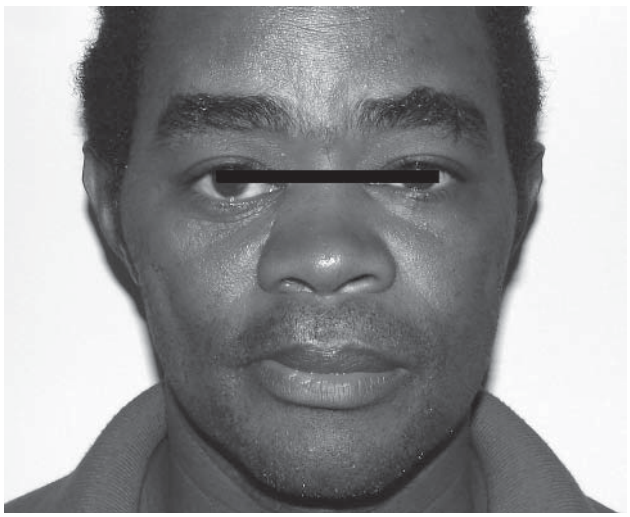

Fig. 10. Little loss of anterior and lateral zygomatic projection and increase in the opening between the eyelids of the right eye in rest position

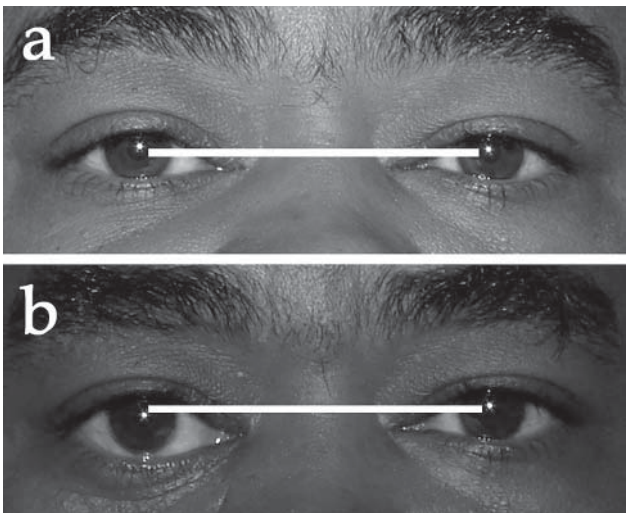

Fig. 11. Changed relation between the pupils, before (a) and after (b) the surgery

The patients who describe their pain as fairly severe, in general belong to the group with soft tissue invasion. The patients who complain of mild to moderate pain generally have intrabony lesions [16]. Rapidly growing odontogenic myxomas are rare, but have been reported in the literature [16,18]. Odontogenic myxomas can be extensive, involving half of the maxilla or mandible including the ramus and the condyle [18,23,24]. The maxillary sinus is often filled with tumor mass that can lead to exophtalmos [29], which did not occur in our case. In the study of Simon et al. [16], with 33 cases, patients with posteriorly located tumors presented late with bigger lesions than those with anteriorly located tumors. This is probably due to more visible disfigurement and disturbance of bite function when the lesions are located in the anterior area.

External and internal cortical displacement can be very evident, with bone destruction and soft tissue protrusion, as seen in the CT of our case, with significant expansion of the maxillary sinus. Our case showed encroachment on the maxillary sinus only. This finding is supported in the literature, where it is reported that maxillary lesions encroach upon the maxillary sinus; however, lesions infiltrating the nasal cavity are rare [23].

It is reported that may occur root resorption although rare, but displacement of teeth is a relatively common finding [4,16,22,24]. Considering this, it is remarkable that Kaffe et al. [18] found root resorption in 9.5% of cases studied and Noffke et al. [17] had presented thirteen cases (of thirty; 43%) with root resorption. Noffke et al. [17] hypothesized that the more frequent occurrence of root resorption in their study may be due to the larger size and subsequent pressure of OMs when compared with those reported in the literature. There was scalloping of teeth in our case, a finding that correlates with the literature [17], and also root resorption, as described by other authors [4,16]. But none was displaced. The lack of tooth extrusion in the case is probably the result of growth into the maxillary sinus.

The microscopic features in this case were compatible with that of odontogenic myxoma, which is comprised of spindle or stellate cells in a mucous material.

It was reported that the size of the lesions correlated with their locularity [18,24]. Multilocular lesions reached sizes larger than 40 mm, thereby exceeding unilocular lesions in the greatest dimension. These findings correspond with the study of Noffke et al. [17]. Our large multilocular lesion also showed that, although there were no small lesions in others patients to compare.

Unicystic lesions were generally smaller in size than the multicystic lesions. Usually, fine trabeculation is found within the myxomas, the border of which can be either well or poorly defined. There seems to be no correlation between the borders of the lesion and internal structure nor between the age of the patient and the size of the tumor. The localization and differences in bony structure also seems to be of importance, since unilocular tumors are mostly located in the anterior parts and the multilocular tumors are found in the posterior parts of the jaws [24].

Some authors had previously associated the OM origin with a myxomatous change of an odontogenic fibroma1 or residual foci of embryonic tissue. However, the odontogenic origin of the neoplasm is supported by its histological similarity to pulpal ectomesenchyme, its exclusive occurrence in close proximity to the tooth-bearing parts of the jaws, occasional association with missing or unerupted teeth, presence of inactive odontogenic epithelium in a minority of cases, and its rare occurrence in other parts of the skeleton, offer support to an odontogenic origin [2].

Odontogenic myxoma frequently displays aggressive infiltration of the adjacent tissue as well as a tendency to recur after surgical removal [23,24]. In order to manage these tumors appropriately, it is imperative to determine their extent.

In addition to conventional films the radiologic examination of cystic jaw lesions can include methods as computed tomography (CT) and magnetic resonance (MR) imaging. These methods have been found to be superior to plain radiographs when establishing the intraosseous extent of the tumor, cortical perforation and soft tissue involvement, and extent [25]. Tooth displacement and root resorption, however, can be observed more reliably on conventional radiographs [25], as showed in the panoramic radiography of our case. It was reported, however, that even with CT examination, several OMs could not be distinguished from other lesions [30]. Magnetic resonance imaging is indicated in cases of suspected recurrent OM, because this modality facilitates the differentiation between fibrous connective tissue and tumor tissue due to different signal intensities [31].

In Brazil, it is still unusual for departments of Oral and Maxillofacial radiology to have their own CT and MR equipment. Thus, examination with these machines usually has to be restricted to high priority cases with: for example, suspicion of malignancy and/or extension into the soft tissues. These equipments are only common in main hospitals with departments of medical radiology.

Despite a considerable number of lesions that may be included in the differential diagnoses of OM, there are some aspects that one must take in consideration, because the myxoma may present variable radiographic features. Unlike giant cell granulomas, multilocular myxomas are often found in the posterior regions and fine trabeculation is seen within the lesions. Giant cell lesion of hyperparathyreoidism can be eliminated if there is no history of kidney disease and serum chemistry is normal. Cherubism, a rare lesion found in children, can be ruled out in most cases because of the age of the patient and clinical findings. In contrast to myxoma, aneurysmal bone cyst presents with symptoms such as tenderness and pain. Intrabony hemangioma can be ruled out because aspiration of a myxoma is nonproductive. The possibility of a malignant tumor has always to be considered. Especially, in older patients the possibility of a metastatic tumor must not be neglected. Radiological features resembling osteosarcoma may be seen. Some cases of Noffke et al. [17] presented with extruded teeth and widened periodontal ligament spaces, mimicking an osteogenic malignancy. A number of lesions should be included in the differential diagnoses; therefore, a biopsy is necessary to ascertain an accurate diagnosis.

Other jaw tumors that can give rise to histological differential diagnostic problems are desmoplastic fibroma and odontogenic fibroma. The former can be distinguished by its compact whorling fibrous fascicles and threadlike nuclei. The latter, odontogenic fibroma, is more fibrous, is encapsulated, and lacks the stellate-shaped cells typical of myxoma [14].

Appropriate surgical and adjuvant therapy depends on a correct diagnosis. Interaction between the clinician and pathologist, especially when dealing with odontogenic lesions, is essential [19]. The current recommended therapy depends on the size of the lesion and on its nature and behavior and can vary from a conservative curettage [3,6,15] to radical excision [26,28]. Complete surgical removal, using curettage and peripheral ostectomy alone is not sufficient as the lesion is not encapsulated and because the myxomatous tissue infiltrates adjacent bone [28]. These characteristics may explain the high rate of recurrence of myxomas, where simple enucleation and curettage alone can have recurrence rates which range from 10 to 33% [4,15,22,29]. Recurrences are the result of insidious local invasion into cancellous bone beyond radiographically visible margins and absent encapsulation [27].

Due to the high reported rate of recurrence of myxomas when treated conservatively, it is the policy of our department that the treatment protocol for OM be a radical primary resection of the tumor with maximal preservation of surrounding anatomic structures, with a peripheral ostectomy of the cavity borders with a large spherical drill, to minimize the recidive chance. This occurs because apparently its clinical and radiological limits may not histologically represent the true limits of the tumor. A conservative surgery based upon enucleation and curettage should be only acceptable for small-sized myxoma. When a small tumor would be detected, the functional and aesthetic result will be less incapacitating even after such radical surgery. It was not our case, where a partial maxillectomy was performed.

Reconstruction can begin immediately following the surgical procedure or delayed until an adequate disease free period has past. Small bony defects (smaller than 5 cm) can be reconstructed using buccal fat pad, or using corticocancellous iliac crest bone graft [32]. Larger defects (more than 5 cm) usually require primary prosthetic reconstruction (obturator) followed by a final obturator, as demonstrated by Leiser et al. [28]. Mandibular lesions can be managed primarily using a reconstruction plate followed by an immediate or delayed vascularized fibular free flap [33], iliac crest graft [34], costochondral graft, or scapular osteocutaneous free flap [35].

According to Leiser et al. [28], usually in large OMs, a disease free period of several years is advised due to the high recurrence rates of the tumor and the morbidity of the donor site. They believe a minimum follow up of 5 year is advisable to establish disease free

status in order to move to the final reconstructive phase of all patients with OM. That is why we decided in the first moment not to reconstruct the defect, and only put placed a titanium mesh to avoid complete collapse of the right cheek. In our case, wide surgical excision led to a good postoperative result without functional sequelae from the ocular point of view and with a small aesthetic deficit that the patient considered not to be important.

CONCLUSIONS

This case highlights the importance of including myxoma in the differential diagnosis of radiolucent radiopaque lesions in the mandible or maxilla. Proper preoperative radiographic examination is important in order to determine the bony involvement and the extent of the tumor. This should include plain radiographs obtained in different projections, and in appropriate cases also three-dimensional techniques as CT-scans and MR-images.

Collaboration of an experienced pathologist is essential, since this diagnosis may lead to either surgery or a more conservative treatment, and the follow-up must be restricted, with imaging exams to detect any early recurrence. In case of large tumor resections, the reconstruction procedures must be delayed until an adequate disease free period has past.

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