

## Case Report

# Odontogenic Myxoma: Presentation of a Case and Literature Review

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### Article History

Received: 05.12.2021

Accepted: 08.01.2022

Published: 27.02.2022

### Journal homepage:

<https://www.easpublisher.com>

### Quick Response Code



**Abstract:** According to the World Health Organization, maxillo-mandibular odontogenic myxoma is a benign tumor of mesenchymal origin. Its frequency, rare etiology, pathogenesis and therapeutic modalities are often discussed in the literature. It is a rare tumor that accounts for 0.41% to 7.19% of maxillo-mandibular tumors. In this present study, we reported a case of odontogenic myxoma of the left jaw that had progressed for several years, in a 25-year-old patient referred to the odontostomatology department of the Idrissa Pouye General Hospital by his attending dentist. The onset of symptoms went back three years. The volume of the mass evolved gradually and slowly over time. The medical examination did not reveal any particular medico-surgical history, or associated general signs. The intraoral examination revealed on inspection a tumor mass occupying the vestibular premolar-molar region of the left maxilla and measuring approximately five centimeters along its longest axis. This mass was bumpy in shape and reddish in color in places. On palpation, the mass was painless, mobile, of firm consistency, not bleeding on contact. Maxillofacial computed tomography revealed the presence of a spontaneously isodense, budding osteolytic process with lobulated contours, blowing the left maxilla in its anterior part. It extended into the hallway and made contact with the inside of the cheek, but did not invade it. The therapy was surgical under general anesthesia. No recurrence was observed at 9 months later. Maxillo-mandibular odontogenic myxoma is a rare, benign and locally invasive tumor. Its odontogenic origin remains the most likely. The clinical aspects or the radiographic presentation are not characteristic and make the differential interpretation difficult. A biopsy is mandatory for the establishment of a final diagnosis. The treatment is surgical. Rigorous monitoring is required during the first two postoperative years, as recurrences are not uncommon.

**Keywords:** Odontogenic myxoma, maxilla, management.

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## INTRODUCTION

Maxillo-mandibular tumor pathology is represented by a group of polymorphic lesions of various origin and nature. It is made up, among others, of non-odontogenic tumors and odontogenic tumors. Odontogenic tumors are tumors associated with the proliferation of epithelial, ectomesenchymal and / or mesenchymal elements originating from the dental apparatus or remnants of tooth-forming tissues. Our study concerns a rare case of odontogenic tumor: odontogenic myxoma [1, 2].

According to the World Health Organization, odontogenic myxoma is a benign tumor of mesenchymal origin. It is a rare tumor that accounts for 0.41% to 7.19% of maxillo-mandibular tumors. Its rare etiology, pathogenesis and therapeutic modalities are often discussed in the literature [3, 4].

The diagnosis can be suspected based on clinical and radiological data, which, however, are non-specific variables. They can often be confused with other benign and malignant lesions. Only histological examination can make the definitive diagnosis with certainty. Surgical excision is the treatment of choice. It should be as conservative as possible, but large enough to reduce the risk of recurrence. These recurrences dominate the therapeutic issue [5, 6].

The reported case is the first documented case of odontogenic myxoma treated in the odontostomatology department of the General Hospital Idrissa Pouye. The objective of this study was to explore the clinical, paraclinical and therapeutic aspects of this rare tumor.

## OBSERVATION

This was a 25-year-old student, referred to the odontostomatology department of the Idrissa Pouye

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General Hospital by a dental surgeon for a left maxillary gingival growth that has been evolving for several years. The onset of symptoms went back three years. The volume of the mass evolved gradually and slowly over time. The medical examination did not reveal any particular medico-surgical history, or associated general signs.

Exobuccal examination revealed facial asymmetry related to a cheek tumor. The teguments were normal, non-inflammatory, without a fistula. The tumor was painless on palpation, leaving the integuments of the cheek free. We noted the presence of left submandibular lymphadenopathy, painless on palpation, firm consistency and mobile compared to the superficial and deep planes (Figure 1).

The intraoral examination revealed on inspection a mass occupying the left maxillary premolar region on the vestibular side and measuring approximately five centimeters along its longest axis. In closed mouth and lips parted, the mass covered the vestibular surfaces of the ipsilateral antagonistic mandibular teeth ranging from 34 to 38. The mass filled the vestibule extending beyond the upper occlusal plane by two cm, without covering the occlusal surfaces. When opened mouth, the mass extended from 22 to the ipsilateral maxillary tuberosity resulting in a palatal version of the upper left molar group. This mass was bumpy in shape and reddish in color in places. Oral hygiene was poor with the presence of tartar on the teeth related to the tumor. On palpation, the mass was painless, mobile, of firm consistency, not bleeding on contact (Figure 2). At the end of the clinical examination, the diagnosis of a benign odontogenic tumor was made. The diagnostic hypotheses of odontogenic myxoma, giant epulis and ossifying fibroma were retained. Radiological examinations were ordered.

The panoramic dental x-ray was not very helpful. However, we could objectify a transparent X-ray image, with more or less clear contours, located at the level of the left jaw. We also noted the presence of retained wisdom teeth (Figure 3). Maxillofacial computed tomography revealed, in a bone window, the presence of a spontaneously isodense, budding osteolytic process with lobulated contours, blowing the left maxilla in its anterior part and invading the left maxillary sinus. It lysed the anterior wall of the left maxillary sinus upon contact and extended into the vestibule making contact with the medial aspect of the cheek without invading it. The lateral extension respected the posterolateral aspect of the maxilla (Figure 4). In the parenchymal window, an inhomogeneous tumor mass was noted in the left maxilla, with the presence of calcification in it (Figure 5).

An incisional biopsy was scheduled and performed a week later. The pathological results of the biopsy favored an odontogenic myxoma of the left maxilla (Figure 6).

The treatment decision was surgical excision under general anesthesia for reasons of comfort and safety given the size of the tumor. The patient was sent for a pre-anesthetic visit for a certificate of fitness. Then a week later, the tumor was removed under general anesthesia. The surgery took place in several stages:

Orotracheal intubation was performed in the patient placed in the supine position. After exo-oral and intraoral disinfection using an iodine solution diluted with 0.9% isotonic saline, we proceeded to the installation of a packing and then a lateral mouth opener. The infiltration of a vasoconstrictor solution was performed to reduce intraoperative bleeding. Equipped with an electric scalpel for the control of hemostasis, an incision of the peri-lesional mucosa, about 5cm long was made, followed by a careful dissection allowing the operating part to be released and resulting in its complete resection (Figure 7 & 8). Careful alveolar curettage with a surgical curette associated with cleaning of the lesion site after resection was performed followed by selective cauterization of suspicious areas. Closure of the operative site was achieved with simple sutures in separate stitches using absorbable suture (Figure 9). The operative part was conditioned and sent for anatomopathological examination (Figure 10).

No complications were noted during the postoperative follow-up. We observed a good evolution during the healing process. Checks have been carried out up to date J1, J7, J14, J30, J60, J90, J120, J180, J270, up to the present day (Figure 11 & 12).



**Figure 1: Extraoral view showing a tumefaction of the cheek leading to left facial asymmetry**

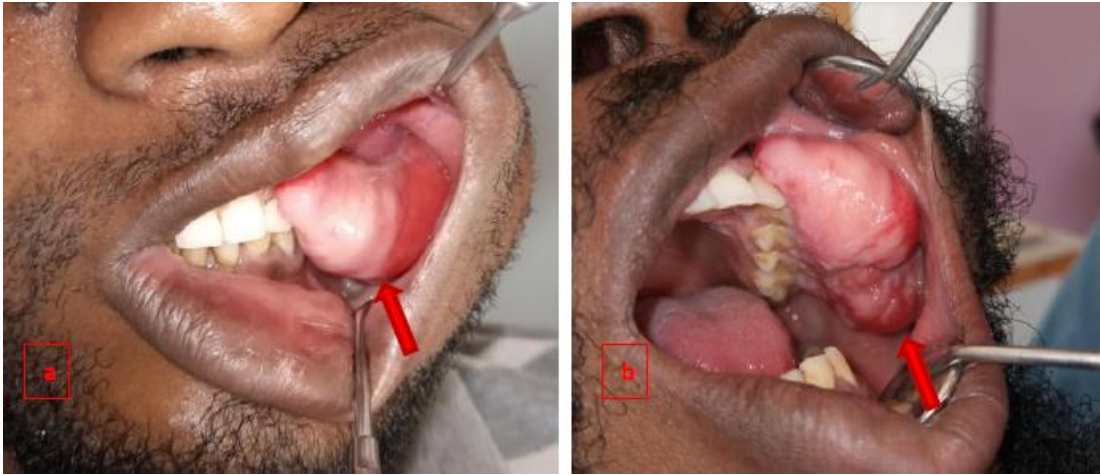


Figure 2: Intraoral view of the odontogenic myxoma of the left maxilla, mouth closed, lips parted (a) and mouth open in (b)



Figure 3: Panoramic dental x-ray of the left maxillary odontogenic myxoma

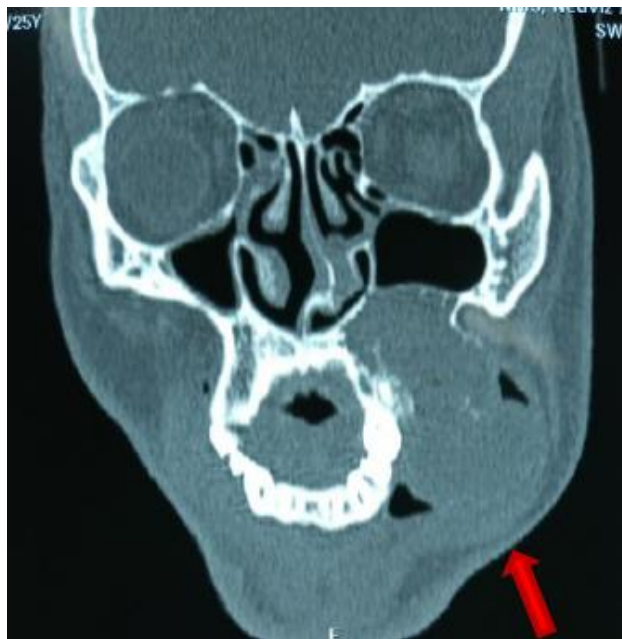
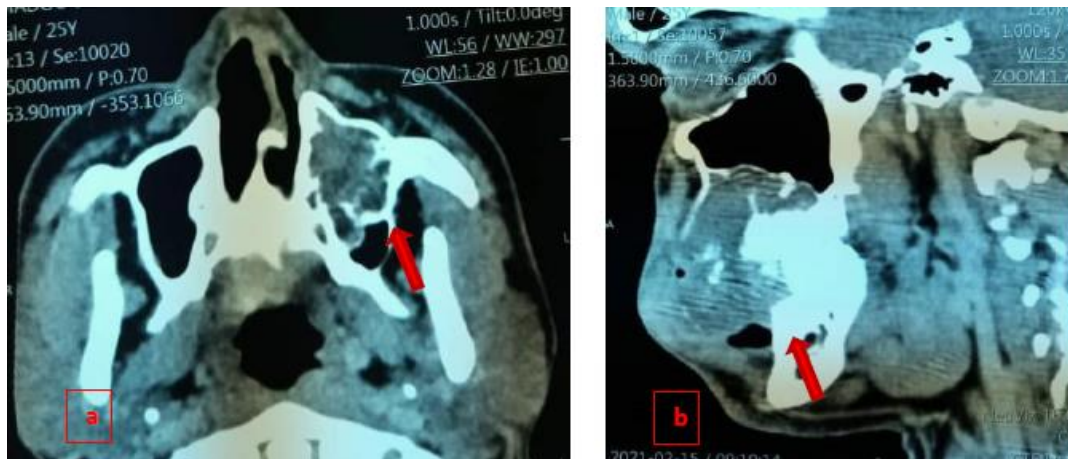
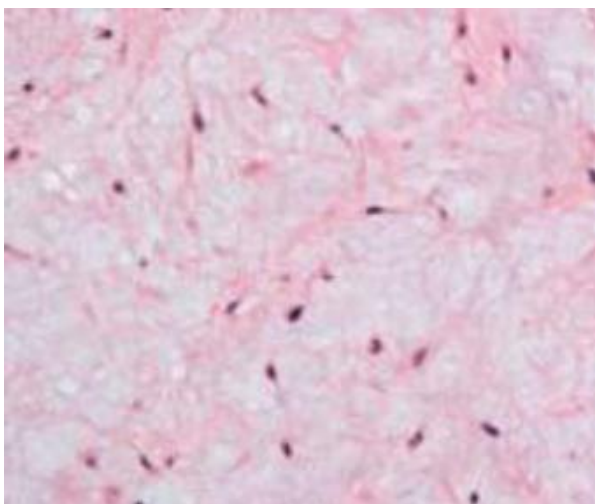


Figure 4: Maxillofacial CT scan, bone window, frontal section of an odontogenic myxoma of the left maxilla



**Figure 5: Computed tomography, parenchymal window in transverse (a) and sagittal (b) section of the left maxillary odontogenic myxoma with invasion of the left maxillary sinus**



**Figure 6: Histological appearance typical of odontogenic myxoma with randomly oriented cells, spindle and round**



**Figure 8: Tumor cavity after excision of the left maxillary odontogenic myxoma**



**Figure 7: Exeresis of the left maxillary odontogenic myxoma, by dissection step by step**



**Figure 9: Sutures in place after excision of the left maxillary odontogenic myxoma**



**Figure 10:** Image of the resected maxillary odontogenic myxoma measuring 08 cm along its longest axis



**Figure 11:** Intraoral view at D270 postoperative



**Figure 12:** Postoperative extraoral view D270 with return of the facial aesthetic profile

## DISCUSSION

Odontogenic myxoma is a relatively rare benign maxillo-mandibular odontogenic tumor. It was first described in 1863, by Virchow [1]. In 1947, Thoma and Goldman distinguished two types of maxillo-mandibular myxoma: central and peripheral [7]. The central myxoma starts in the bone, proliferates within this bone, blows and deforms the bone cortices. In the extreme, it perforates the cortical bone and spreads into the soft tissue. Peripheral myxoma is most often found in an alveolar process. An osteolytic reaction underlying this fibromucosal lesion may appear later [8, 9]. Soft tissue myxoma is rarer than intraosseous myxoma [4].

Many authors consider myxomas of the orofacial sphere as specific lesions of the maxillae and mandible, without any direct comparison with extrafacial myxomas [1, 7].

The pathogenesis of odontogenic myxoma is controversial in the literature, several hypotheses being advanced. Cahn et al, hypothesized that the maxillo-mandibular myxoma was a dysplastic condition arising from the osteogenic mesenchyme, close to fibrous dysplasia; it would be a myxoid variant with a greater immaturity of the mesenchyme, without osteogenic properties [10]. However, the most accepted hypothesis according to several authors, supposes that the myxoma derives from components of the mesenchymal tissue of the dental germ, either at the level of the dental papilla, or at the level of the follicle, or again at the level of the periodontal ligament. Myxoma is believed to be a rare benign maxillo-mandibular odontogenic tumor of mesenchymal origin. Odontogenic tumors derive from elements of odontogenesis with aspects of varying tumor maturation at the stages of organogenesis of a normal tooth [5, 8, 11]. The arguments in favor of an odontogenic origin are the localization only in the bones of the facial skeleton, the site in the dentate region, mainly molar and the frequent association with certain dental anomalies (malposition, agenesis) [5, 8, 12].

Odontogenic myxoma represents 3 to 6% of odontogenic tumors and 0.41 to 7.19% of maxillo-mandibular tumors. [3, 4, 13]. In Asia, Europe and America, relative frequencies between 0.5 and 17.7% have been reported. It is the second most common odontogenic tumor after ameloblastoma in Africa with relative frequencies between 1 and 19% [9, 11]. Its incidence is very variable according to the authors, ranging from 0.04% to 2.3% [13].

The distribution by age and gender also varies according to the authors and the study sample. It is rare to see it before 10 years and after 50 years, the average age of onset being 20 to 30 years [11, 13, 14]. In a study by Simon et al, the youngest patient was three months old and the oldest was 64 years old. [15]. For some authors, women are more often affected than men. [5,

11]. However, other authors have not observed a gender difference [4, 14]. In our study the patient was 25 years old and male.

Concerning the distribution of the myxoma at the level of the bone bases, the mandibular localization is slightly more frequent than the maxillary one. The posterior maxillo-mandibular sectors are most often affected [5, 9, 11]. In this study, the tumor lesion was located in the left maxilla.

Odontogenic myxoma often goes unnoticed with an incidental discovery during a routine clinical examination. The consultation would be more motivated by the presence of a facial asymmetry due to a unilateral bone tumor, painless and without functional disorders. The consultation time according to the literature varies between one month and 08 years [8, 14]. In our case, left cheek tumefaction associating facial asymmetry was the reason for consultation.

Clinically, the intraosseous lesion is manifested by a swelling which is usually painless and slowly growing, which can lead to facial asymmetry. However, some authors have reported symptoms such as pain and paresthesia [11]. Palpation reveals a mass of firm consistency in the case of a central form, which may be elastic when there is an invasion of the cortical or in the event of a peripheral form, as in our case with the invasion of the anterior wall of the left maxillary sinus showing an elastic lesion [4, 14].

In the maxilla, in particular, the intrasinus development of odontogenic myxoma leads to an asymptomatic extension for a long time, which can lead to complete invasion of the maxillary sinus and / or an often late discovery. It should be noted a possible invasion of soft tissues in the event of destruction of the cortices [11, 16].

Often associated dental signs are the appearance of dental mobility, root resorptions, displacement or loss of teeth. Usually with tooth mobility, pulpal vitality is not compromised [11, 16].

Several imaging techniques provide information on the characteristics of the tumor as well as its relationship to neighboring structures. In first intention, the orthopantomogram or dental panoramic X-ray will be carried out. It makes it possible to specify the anatomical relationships and the intraosseous development of the tumor [14]. Computed tomography (CT) and more rarely, magnetic resonance imaging (MRI), may also be requested from the outset in the particular case of certain large tumors, for an accurate lesion assessment [17]. Computed tomography is very informative, allowing an accurate analysis of the integrity of the bone walls. It also helps reveal the existence of septa within the lesion. CT is essential to determine the modalities of the surgical intervention

based on the three-dimensional topography of the lesion and neighborhood relationships [14]. MRI can more accurately determine the extent of the lesion, especially in soft tissue [13]. On MRI, odontogenic myxomas have a variable signal: an odontogenic soft tissue myxoma gives a hypersignal in T2 and a hyposignal in T1; for intraosseous odontogenic myxoma, the reverse is true [4]. In our study as radiological examinations we requested a panoramic dental x-ray followed by a maxillofacial computed tomography.

The classic radiological appearance of myxoma is that of a radiolucent image in the bony, polygeodic region with relatively sharp and distinct polycyclic edges. Sometimes radiopacity may be associated due to the presence of mineralized tooth tissue. The confluence of the geodes forms partitions which intersect at sharp or right angles thus giving gaps, of variable size, a more polygonal configuration than rounded often described in the literature under the name of honeycombs, soap bubbles or tennis rackets. Myxoma may appear as spicules resulting from tumor breaking through the cortical bone, invading surrounding soft tissue [8, 12]. Myxoma can also appear as a monogeodic or unilocular cystic lesion. These osteolytic lesions are often surrounded by a marginal area of high radiological density [5]. The anomalies of the dentition found are mainly represented by rhizalysia, dental displacements and / or alveolysis [17].

The macroscopic appearance of this tumor is characteristic. The tumor has a lumpy, bumpy, or multi-lobed appearance. It is well defined, shiny, smooth, whitish, greyish or yellowish and little hemorrhagic on the surface [8]. The tumor volume varies in the literature from 05 to 95 mm. It is often more important than the radiological appearance would suggest. Its consistency depends on the abundance of collagen fibers and is highly variable, but homogeneous. It can be elastic, gelatinous (myxoma) or firm (fibromyxoma) [8, 14, 16].

The microscopic appearance of this tumor reveals two types of stellate and spindle-shaped cells, with scanty basophilic cytoplasm, with a small hyperchromatic nucleus. Cell density is variable. Ground substance is the most abundant element; loose stroma, "myxoid-like", is compared to the primitive mesenchyme of the tooth-germ stellate reticulum. We note the presence of non-secreting, round and dark cells, and spindle-shaped, triangular or stellate cells, whose long fibrillar-like extensions tend to intersect [8, 26].

The vessels are absent. Islets of dental epithelium, apparently inactive can be observed, which proves odontogenic origin according to some authors [18, 19]. Collagen fibers are scarce, except in the case of myxofibromatous varieties. There is no cell atypia and mitotic activity is low, which is compatible with the

slow growth of the tumor [5, 14]. At the periphery, myxomatous tissue enters the trabecular spaces, producing islets of residual bone. This aspect explains the difficulty in eliminating the lesion in a conservative manner [11].

Histochemical and ultrastructural study reveals that tumor tissue is rich in acidic mucopolysaccharides (80% hyaluronic acid and 20% chondroitin sulfate) [5]. In immunohistochemistry, myxoma tumor cells express vimentin. Smooth muscle actin is classically positive for all tumor cells. For the S100 protein (marker of nerve differentiation), the immunostaining of tumor cells is variable but classically weak and focal or simply negative [3, 8].

The diagnosis of myxoma is generally suspected based on clinical and radiological data, but it is the pathological examination that can make the diagnosis with certainty. It is the pathological examination of the excisional piece that provides the definitive diagnosis.

Clinically, the differential diagnosis of odontogenic myxoma can be made with other lesions of the reaction / inflammatory or tumor type (benign and / or malignant) of the oral cavity such as: epulis, giant cell granuloma, ossifying fibroma, ameloblastoma and osteosarcoma [4, 5, 8, 11].

The epulis is a fairly characteristic hyperplastic pseudotumor or reactive hyperplasia, usually starting from the sulcus or inter papillary, protruding on the gum tissue. It classically sits on the vestibular side of the maxillary incisor-canine region [20, 21, 22, 23]. Central giant cell granulomas affect the anterior sector of the maxilla more frequently than the posterior one [5, 11, 13]. Ossifying fibroma clinically manifests as a mass of firm consistency with a sharp borderline. It sits mainly on the palate or on the retro-molar trine. The presence of collagen fibers which characterize the varieties of fibromyxomas can be confused with certain fibroids at the histological level [5, 11, 13]. Ameloblastoma is usually found in the area of the third molars, especially in the mandible. Odontogenic myxoma is regularly found throughout the dentate portions [5, 11, 13]. Bone sarcomas can be evoked by the rapidity of progression and the lytic aspects with burning cuts of grass. Histologically, the presence of certain nuclear atypias in the myxoid component can suggest bone sarcoma (osteosarcoma or chondrosarcoma) and make the diagnosis difficult [5, 11, 13]. In our study, the diagnostic hypotheses of giant epulis, ossifying fibroma and odontogenic myxoma were retained.

Myxoma is mainly treated by surgical means. Although the histological appearance remains benign, the risk of recurrence dominates the therapeutic issue. Two different surgical therapeutic approaches are found

in the literature, conservative treatment and radical treatment [5, 8, 14, 16]:

- Conservative treatment consists of enucleation and careful curettage, with or without chemical or electrical cauterization of the walls of the excisional cavity. It often turns out to be difficult due to the soft, gelatinous nature of the tumor tissue
- Radical treatment includes an interrupting or non-interrupting bone resection, with safety margins between 0.1 to 1.5 cm from the edges of the lesion. This treatment often results in loss of bone substance and / or soft tissue requiring immediate or deferred reconstructive surgery or prosthetic restoration.

In general, the size of the tumor and its location determine the indication for surgery. However, the patient's age and medical history may also influence the choice of treatment [14].

• **Tumor size:**

Large lesions are traditionally approached extraoral. This approach, however, leaves the patient with an external facial scar [8, 9]. The intraoral approach is suitable for small lesions. Conservative treatment with enucleation or enucleo-resection is recommended for a non-extensive lesion with direct view allowing easy monitoring of healthy bone margins. Many authors agree on the value of enucleation, associated with intensive curettage, with or without chemical or electrical cauterization of the walls of the tumor cavity for small tumors [15, 24].

Due to the significant aesthetic and functional sequelae, radical treatment is reserved for large or recurrent lesions [14]. Because of the local aggressiveness and the high risk of recurrence of odontogenic myxoma, some authors consider radical treatment to be the treatment of choice for this tumor [25, 26].

• **The location of the tumor:**

Slootweg and Wittkamp in 1986 suggest that it is easier to remove all visible tumor tissue in the mandible by careful curettage. However, still according to these authors, in the maxilla, the proximity of vital structures and the greater risk of diffusion rule out the conservative approach because of the greater risk of recurrence. More radical approaches are recommended in these cases [27]. In certain specific cases of large intraosseous tumors located near noble structures, tumor resection may require an interrupting osteotomy [8, 9, 17].

Conservative treatment should be performed with enucleation and curettage in individuals who prefer this treatment, or in those whose anesthetic risk is too great to undergo radical surgery [9, 11].

For Marx and Stern, curative treatment, a term used to indicate radical surgery for an odontogenic myxoma, is completed by the resection of 1 to 1.5 cm of healthy bone [28]. It should be noted that the peripheral myxoma is well limited and non-invasive unlike the central myxoma, its excision remains conservative [4]. In our case, the choice of conservative treatment was made after discussion with the patient and overall consideration of the situation (first intervention, the patient's age, and most importantly, respect for his choice).

Odontogenic myxoma is a benign tumor. No case of metastasis or malignant transformation has been reported in the literature. Its cell proliferation index was found to be low and it was therefore suggested that the invasiveness, and its tendency to recur, would result from the dissemination of the tumor tissue in the operative field due to the gelatinous consistency and ill-defined boundaries [9]. The recurrence rate varies according to the series but remains high; on average it is 25%. [8, 14]. Recurrences after conservative treatment are twice more frequent for maxillary than mandibular lesions with a respective percentage of 80.9% and 31.2% [5]. The time to recurrence varies in the literature between two years and 15 years, [29, 30]. However, it has been noted in the literature that some recurrence can appear much later, up to 30 years after the initial surgery [31].

Long-term follow-up is a necessity. Clinical and radiological monitoring should be carried out regularly and for a long period of time for at least the first three years following surgical treatment [11, 14]. It should be followed closely every three months for the first year, then every six months for two years and finally, annually for several years thereafter. These first three years correspond to the period during which the neoplasm is most able to reproduce [11, 13, 14].

## CONCLUSION

Maxillo-mandibular odontogenic myxoma is a rare, benign and locally invasive tumor. Its odontogenic origin remains the most likely. Odontogenic myxoma represents 3 to 6% of odontogenic tumors. Its distribution according to age and sex differs according to the authors. The clinical aspects or the radiographic presentation are not characteristic and make the differential interpretation difficult. A biopsy is mandatory for the establishment of a final diagnosis. The treatment is surgical. Rigorous monitoring is required during the first two postoperative years, as recurrences are not uncommon.

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**Cite this article:** Diatta M, Kwedi K. G. G, Kane M, Gassama B. C, Kounta A, Ba A, Tamba B, Niang P. D, Dia Tine S (2022). Odontogenic Myxoma: Presentation of a Case and Literature Review. *EAS J Dent Oral Med*, 4(1), 48-56.