

Case Report

Odontogenic Myxoma in the Anterior Part of the Mandible—A Case Report

Monika Burja Vladić ^{1,*} , Marko Vuletić ^{2,3} , Sven Seiwerth ⁴ and Dragana Gabrić ^{2,3} ¹ Health Center Metlika, 8330 Metlika, Slovenia² Department of Oral surgery, School of Dental Medicine, University of Zagreb, 10000 Zagreb, Croatia³ Department of Dental Medicine, University Hospital Centre Zagreb, 10000 Zagreb, Croatia⁴ Department of Pathology, School of Medicine, University of Zagreb, 10000 Zagreb, Croatia

* Correspondence: mburja@sfzg.hr

Abstract: Odontogenic myxoma (OM) is a rare, benign intraosseous tumor that arises from the dental follicle, dental papilla, or periodontal ligament. It typically affects the mandible, maxilla, and sinuses, more frequently in women than men. The histopathology of OM is characterized by a myxoid stroma with varying degrees of collagenization and spindle cells. Treatment is surgical, but there are no standardized guidelines. Available options include enucleation and curettage, with conservative approaches offering better aesthetics and functionality but a higher recurrence risk. This case report aims to discuss a rare presentation of OM in the anterior part of the mandible of a 22-year-old female patient with Hashimoto's thyroiditis and idiopathic thrombocytopenic purpura. The patient presented with painless swelling and tooth mobility in the symphysis region. Radiographic evaluation revealed a unilocular lesion extending from the left second incisor to the right first incisor, with no root resorption. Histopathological analysis confirmed the OM diagnosis. Due to the patient's young age and the size of the lesion, a conservative treatment approach was chosen, with the potential for future implant-prosthodontics rehabilitation. The case underscores the importance of thorough clinical, radiological, and histopathological assessment to ensure accurate diagnosis and management of OM. Long-term follow-up is essential due to the increased risk of recurrence in younger patients.

Keywords: odontogenic tumors; Hashimoto disease; mandible; curettage; surgery; case report



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1. Introduction

Odontogenic myxoma [OM] is a rare intraosseous lesion of a benign nature, with a tendency for local aggressiveness. This type of tumor originates from the primitive mesenchymal tissue of the dental follicle, dental papilla, or periodontal ligament [1].

OM was first mentioned by Thoma and Goldman in 1947 [2], and, according to the literature, it accounts for 1.9% to 6.3% of all odontogenic tumors [3]. The latest classification of the World Health Organization [WHO] from 2022 classifies OM as the third most common intraosseous tumor, occupying 3–6% of all types of odontogenic tumors, after ameloblastoma and odontoma.

The prevalence of this condition is higher in woman compared to men [1.5:1], typically occurring between the ages of 20 and 30 [3]. As of yet, the existing research has not identified any correlation with syndromic diseases or other tumors [4].

In addition to its occurrence in adulthood, odontogenic myxoma can also manifest in the pediatric population. However, its incidence is lower and recurrence rates are

comparatively lower than those observed in adults, but it can also lead to notable bone abnormalities [5].

Besides the term “myxoma”, the term “fibromyxoma” is also often used, which is more appropriate when the collagen fibers on the pathohistological findings are more prominent [6].

The etiology of odontogenic myxoma remains elusive. According to Tavakoli and Williams [7], OM exhibits the highest degree of microscopical and biochemical similarity to Wharton’s mucus, which is typically present in the fetal umbilical cord. The latest 2022 World Health Organization classification of odontogenic tumors [3] states that the most important differential diagnosis of OM is a developing dental papilla or a normal/hyperplastic dental follicle that is histologically almost identical to OM, and the final diagnosis is based on clinical and radiological features. The classification also states the pathohistological characterization in the form of stellate and spindle cells dispersed in an abundant myxoid extracellular matrix, which also contain odontogenic epithelium.

Although most authors point out that the tumor is more often localized in the mandible, especially in the premolar and molar region, the tumor can also affect the maxilla, especially in the sinus area [8–13]. Typically, it is unintentionally found or only after it has reached a substantial size, leading to facial abnormalities [14]. The pathohistological features of the condition involve a myxoid stroma that exhibits varying levels of collagenization and contains spindle cells [3].

A standard clinical presentation entails a painless enlargement accompanied by cortical expansion and tooth displacement [15]. The radiological findings can vary from a unilocular to a multilocular lesion, with well-developed bony trabeculae within the lesion described in the literature as honeycomb, soap bubble, or tennis racket net [16].

In the differential diagnosis, numerous other conditions that can radiologically overlap with OM should be excluded, such as ameloblastoma, intraosseous hemangioma, aneurysmal bone cyst, glandular odontogenic cyst, central gigantocellular granuloma, cherubism, odontogenic keratocyst, osteosarcoma, and metastatic tumors, and the final diagnosis should be based on the clinical, radiological, and pathohistological picture [17].

Surgical treatment is an option for the treatment of this tumor, but there are currently no standard guidelines. Different treatment modalities can be used—from enucleation and curettage to more radical ones, such as *en bloc* resections.

A more conservative approach is usually associated with a higher recurrence rate, but also with a better quality of life in terms of aesthetics and functionality [7,13,16].

The purpose of this paper is to present a rare example of odontogenic myxoma in the anterior part of the mandible that crosses the midline in a patient suffering from idiopathic thrombocytopenic purpura and Hashimoto’s thyroiditis, with a review of the clinical, radiological, and histological features of odontogenic myxoma and its differential diagnosis.

2. Case Report

A 22-year-old woman came to the Department of Oral Surgery of the Clinical Hospital Center Zagreb due to swelling in the area of the symphysis and mobility of the lower incisors. No complaints other than swelling were reported. The patient reported having Hashimoto’s thyroiditis and idiopathic thrombocytopenic purpura in the medical history.

Clinical examination revealed that skin of the affected area was of normal color, as was the intraoral mucosa covering the symphysis of the mandible (Figure 1).

Radiological analysis with cone beam computed tomography (CBCT) and an orthopantomogram showed a unilocular, well-defined, expansive radiolucent formation, extending from the second left incisor and, crossing the midline, to the first right incisor (Figure 2). The lesion visibly spread to the incisor roots, without resorption.



Figure 1. Clinical examination.

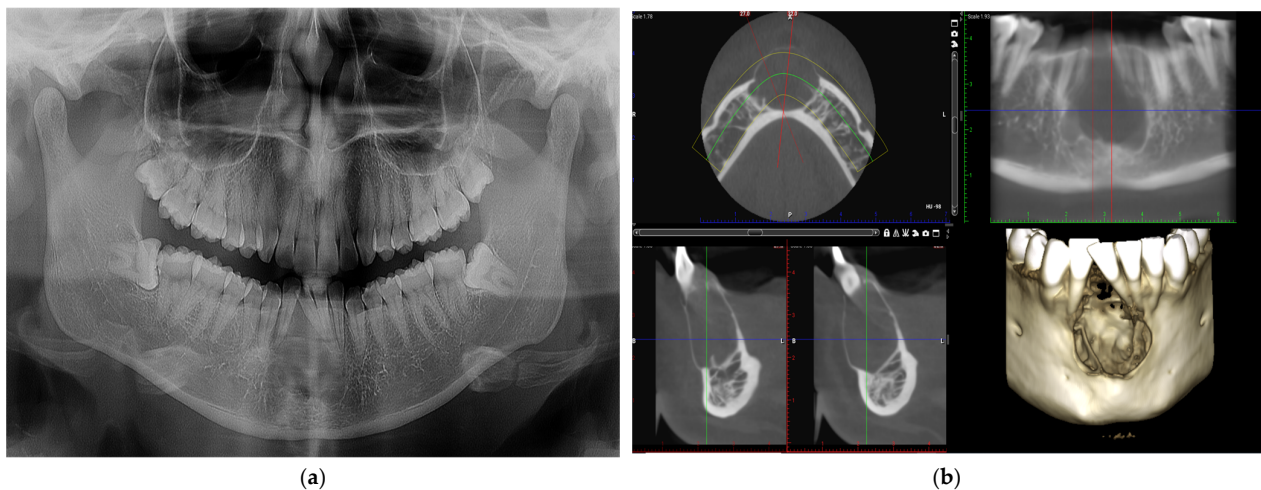


Figure 2. (a) Preoperative panoramic radiograph; (b) CBCT scan.

Under local anesthesia (Ubistesin 3M Deutschland GmbH, Neuss Germany, 3×1.7 mL), an incisional biopsy of the change was performed. The wound was sutured primarily.

The pathohistological finding showed that it was a formation made of a myxoid matrix with areas of somewhat stronger presence of collagen, in which cells were predominantly spindle-shaped with darker nuclei, with locally present anastomosing of elongated cytoplasm. The finding corresponded to myxoma of the gnathic bone (Figure 3).

Afterward, due to the clinical and radiological finding and patient's age, a procedure with enucleation and curettage under general anesthesia was planned.

A crestal incision and two vertical releasing incisions were made in the anterior mandible, and the full mucoperiosteal flap was raised to expose the alveolar ridge and demonstrated an inflated bone with partial fenestration of the tumor formation. The affected teeth were extracted, and a complete enucleation of the tumor was performed with curettage of the surrounding bone until reaching a healthy one (Figure 4).

To promote better soft tissue healing, plasma rich in growth factors or PRGF (Endoret, BTI, Vitoria-Gasteiz, Spain) fraction F2 and PRGF membrane fraction F1 were prepared according to the manufacturer's procedure and inserted into the wound and sutured with 3–0 absorbable sutures (Figure 5).

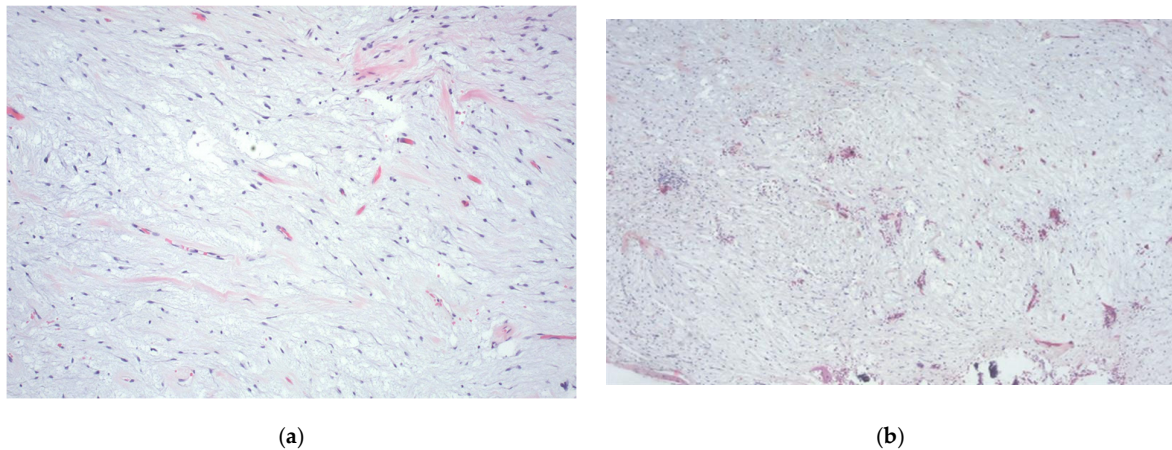


Figure 3. (a) Hematoxylin and eosin staining (magnification $\times 10$), (b) hematoxylin and eosin staining (magnification $\times 4$). From the histopathological report: A whitish specimen measuring $3 \times 2 \times 5.2$ cm was received, processed using step-sectioning. Histologically, the lesion consisted of a myxoid matrix with areas of slightly more pronounced collagen presence. Spindle-shaped cells with darker nuclei, embedded within the matrix, were predominant and occasionally showed anastomosing elongated cytoplasmic processes. No atypia was observed, and mitotic activity was low. Fragments of compact bone were also present in the tissue. A thin, delicate connective tissue capsule was observed over most of the lesion's surface, although it was absent in certain areas, where extravasated erythrocytes were noted. The findings were consistent with a myxoma of the gnathic bone. It could not be conclusively determined whether the described lesion was excised completely from the jaw.

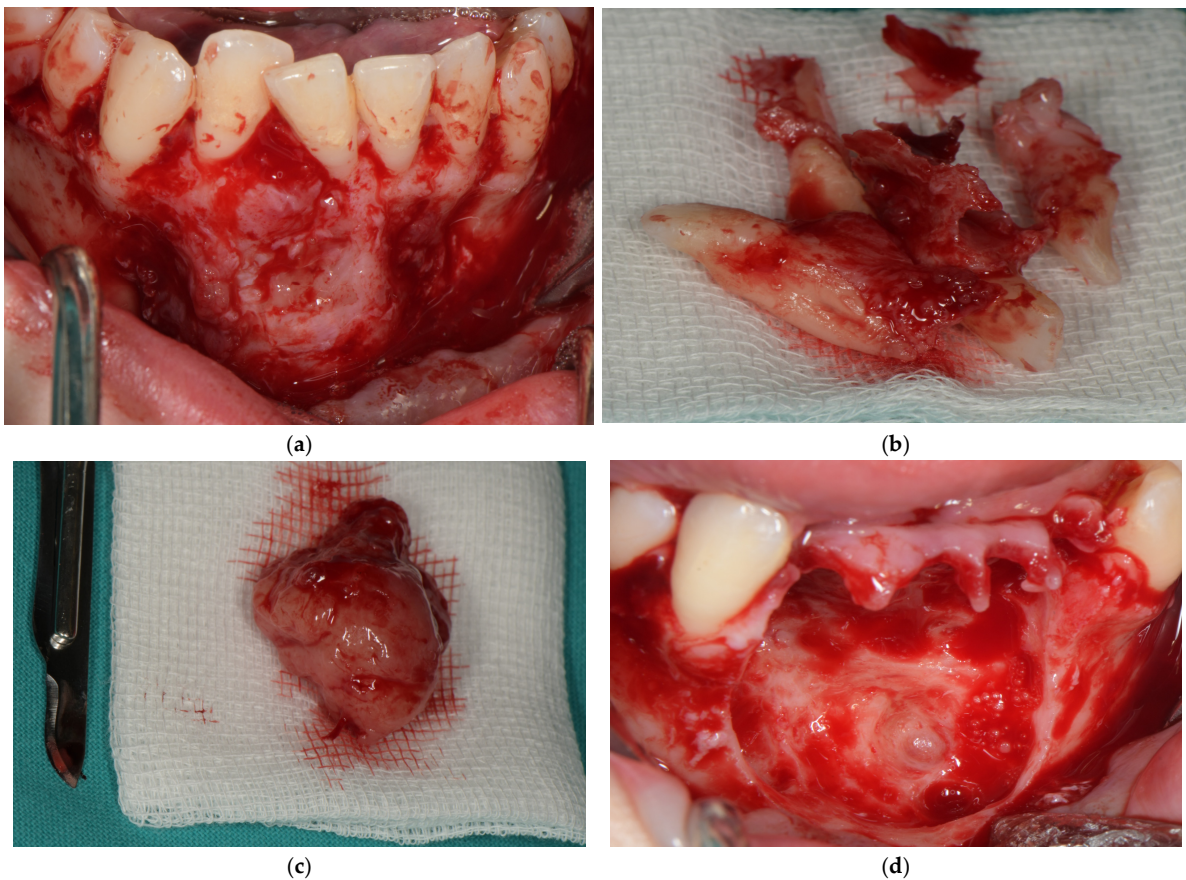


Figure 4. (a) Raised mucoperiosteal flap showing inflated bone; (b) extraction of the affected teeth; (c) gross examination of the OM; (d) bone after completion of enucleation and curettage.

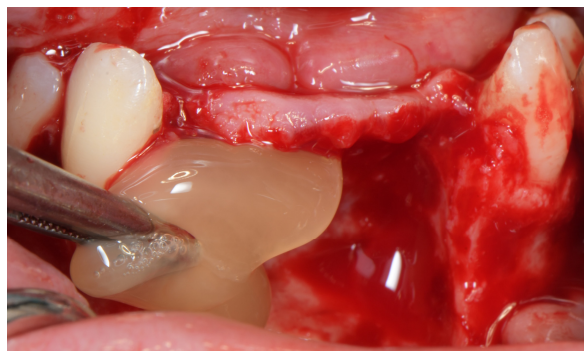


Figure 5. Fibrin membrane (F1) and fraction F2.

To avoid postoperative complications, antibiotic therapy (Klavocin bid 875 mg amoxicillin + 125 mg clavulanic acid, Pliva, Zagreb, Croatia), was initiated twice daily for seven days postoperatively, and 0.12% chlorhexidine mouthwash and oral analgesics were also prescribed.

The healing period went without reported problems. The patient has been under follow-up care for a year and a half, with regular controls, radiological imaging, and no signs of a reoccurrence (Figure 6).



Figure 6. Postoperative panoramic radiograph.

As a temporary solution, the patient was provided with an *essix* splint (Figure 7). In the future, when complete bone regeneration is observed on a routine follow-up orthopantomogram, and there are no clinical or radiological signs of potential recurrence, implant-prosthetic rehabilitation will be performed in agreement with the patient. The time required for this process is individualized.



(a)



(b)

Figure 7. (a,b) *Essix* splint temporarily replacing lost frontal teeth.

3. Discussion

Ten case reports of OM from the last five years were discovered by searching the literature for “odontogenic myxoma” in the PubMed database. Only two of the cases listed were set in the maxilla; the majority of cases were found in the mandible” it should be written: “Most authors report the finding of odontogenic myxoma predominantly in the mandible, in the premolar and molar region [1,12,18]. Ten case reports from the last five years were discovered by searching the literature for “odontogenic myxoma” in the PubMed database. Only two of the cases listed were set in the maxilla [19,20]; the majority of cases were found in the mandible [7,21–25].

In our study, the lesion was present in the anterior part of the mandible, including the front teeth. Most often, OM affects the tooth-bearing areas, but it can also be found in the area of the condyle of the mandible [1,26] and in the area of the gingiva [27–29]. For these rare variants, the nomenclature “peripheral odontogenic myxoma” has been suggested by Raubenheimer and Noffke [29]. OM in the maxilla region is less common, although certain authors suggest that it exhibits greater aggression compared to the mandible, particularly where it intersects the maxillary antrum [16].

Although considered a benign tumor, OM often has a locally aggressive effect on the surrounding bone [16]. However, unlike the present case, OMs usually do not tend to spread across the midline. By searching the literature, we came across only a few cases where the OM crossed the central line [30,31]. Differential diagnosis should take into account conditions in which the spread of the lesion across the midline is a more common finding, such as central gigantocellular granuloma [32], osteosarcoma [33], fibrous dysplasia [34], aneurysmal bone cyst [35], or radicular cyst [36,37]. The tumor rarely causes root resorption and is most often found between the roots [14], as in the present case. Due to the potential overlap between radiological findings and the mentioned entities, relying just on radiological diagnosis is inadequate for the definitive diagnosis of OM, and clinical and pathohistological features should also be considered [14,18,25].

Most odontogenic myxomas are almost always asymptomatic, with the presence of swelling. Nevertheless, some individuals may feel pain while chewing or biting when the tumors infiltrate nearby structures. This may eventually result in neurological symptoms, such as hypoesthesia, paresthesia, dysesthesia, and facial asymmetry [21,24,38]. Similarly, no complaints other than swelling were reported by our patient.

In the presented case, pathohistological findings revealed myxoid matrix with areas of more strongly expressed collagen with spindle-shaped cells, which is in line with the current definition of OM in the WHO classification [3]. Other authors [19–21,23,25] reported similar pathohistological findings.

From a radiological perspective, the lesion may exhibit either unilocular or multilocular radiolucency, characterized by well- or ill-defined, diffuse boundaries [11]. Most of the authors [7,20–22,24,25] reported multilocular appearance with buccal and/or vestibular cortex destruction in the case of the affected mandible. In our case, radiological analysis revealed a unilocular radiolucent lesion. Considering the frequent radiological variability that often causes diagnostic dilemmas, Zhang et al. [39] described the six most common radiological manifestations using conventional panoramic images. The results of their research showed that multilocular type II is most often seen, which are described as multiple radiolucent areas of different sizes, separated by bony trabeculae.

To the best of our knowledge, we found no connection between OM and other tumors in the literature [4]. Therefore, the simultaneous occurrence of OM and Hashimoto’s thyroiditis and idiopathic thrombocytopenic purpura is most likely a coincidence. In addition, it is rare for myxoma to appear in places other than the jaw. In the study of more

than 6000 bone tumors, McClure and Dahlin [40] found only three myxomas that were not localized within the mandible and maxilla.

OM is associated with potentially aggressive behavior and high recurrence rates averaging 25%, therefore the primary therapy is surgical. However, the choice between conservative treatment in the form of enucleation and curettage or radical surgical therapy, which includes segmental or block resection with partial mandibulectomy, is still controversial and there are currently no standardized guidelines for the treatment of OM [41].

Although conservative treatments are significantly less invasive, the risk of recurrence after such therapy is higher due to the lack of encapsulation of the tumor itself. In addition, myxomatous tissue infiltrates the surrounding bone tissue, thus increasing the possibility of recurrence as well. [42].

Several researchers have recommended tumor size as the main determinant for radical or conservative surgery. The same authors believe that recurrence is directly related to the type of therapy, and the rate after simple enucleation and curettage was reported to be as high as 25% [43]. Boffano et al. [44] suggest enucleation and curettage in the case when the diameter of the tumor is less than 3 cm, and segmental resections are recommended in the case of larger tumors. Chrcanovic et al. [12], in their study including 1692 patients, reported that curettage had the highest rate of recurrence, while marginal and segmental resection had the lowest rate of recurrence. Also, according to the results of above-mentioned study, enucleation and peripheral osteotomy showed better results in the recurrence rate rather than enucleation alone or in combination with curettage. Saalim et al. [45] reported a 19% OM recurrence rate after conservative treatment [11-year follow-up], and after resection, the recurrence rate was 6% [9-year mean follow-up]. The conclusion of their study is that there is no statistically significant difference in the recurrence rate between conservative and resective treatment. In addition, they recommend a more conservative treatment approach whenever possible, in order to preserve the patient's quality of life.

Lifelong follow-up is also advised, as it has been documented that OM can recur even after 30 years [14]. Having in mind the patient's age and quality of life, a decision of a more conservative treatment in the form of enucleation and curettage was performed, in order to leave the option of reconstruction. In order to reduce postoperative complications, PRGF was used.

PRGF is a standard protocol at our clinic, which has been proven to be effective at the postoperative period and for soft tissue healing resulting in reduced postoperative pain and swelling. Growth factors in PRGF aid in initiating the migration of undifferentiated stem cells to the site and stimulate their proliferation [46]. In addition, PRGF might promote the growth and differentiation of mesenchymal stem cells (MSCs), potentially enhancing bone healing [47]. Preoperatively, the patient was prepared from a hematological standpoint with additional doses of platelets. Considering her baseline platelet levels, her thrombocytopenia did not pose a limitation for oral surgical procedures. So far, we have not noticed that Hashimoto's limits our use of PRGF.

Follow-up after a year and a half did not show recurrence. The limitation of this case report is a shorter follow-up period compared to the previously published studies.

Currently, there is a lack of standalone pharmaceutical treatment or supplementary interventions to surgical therapy. However, Pereira et al. [48] report on the activity of the MAP/ERK pathway [English *mitogen-activated protein kinase/extracellular signal-regulated kinase*] in odontogenic myxoma, which contributes to tumor development. In *in vitro* conditions, with a special inhibitor of the MAP/ERK pathway, the researchers managed to reduce the tumor and its spread. The results represent the basis for further targeted research on potential "target" pharmacological therapy in the treatment of OM.

4. Conclusions

When establishing the diagnosis, it is imperative to consider the clinical, radiological, and histological findings. Certain attributes of OM exhibit similarities with both benign and malignant tumors. The treatment plan should consider the patient's age and gender, as well as the specific location and dimensions of the lesion. Occasionally, radical reconstructive surgery is the preferred therapeutic option, but it should be postponed until sufficient monitoring is conducted to eliminate the possibility of occurrences.

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Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: Data are contained within the article.

Conflicts of Interest: The authors declare no conflicts of interest.

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