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Case Report

# A case of a rapidly expanding odontogenic myxoma of the mandible

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ABSTRACT

Odontogenic myxoma (OM) is a rare benign neoplasm with locally aggressive behavior. We present an unusual case of OM as a rapidly expanding lesion in the mandible of a 62-year-old woman who underwent segmental mandibulectomy and primary mandibular reconstruction using a titanium plate. Histologically, we observed stellate and spindle-shaped cells lying loosely in abundant mucoid stroma. Immunohistochemical staining was positive for smooth muscle actin and vimentin and negative for desmin and p53. The patient was followed up for 24 months without a recurrence. Long-term follow-up is required due to the high risk of recurrence.



## 1. Introduction

Odontogenic myxoma (OM) is a rare benign neoplasm of the jaws that accounts for 1–17.7% of all odontogenic tumors [1–3]. Thoma and Goldman first described OM of the jaws in 1947 [4]. This tumor is asymptomatic and slow growing, and characterized by stellate and spindle-shaped cells embedded in an abundant myxoid or mucoid extracellular matrix [5]. OM develops mostly during the second and third decades of life and is preferentially located in the mandible [2,3,6–13]. In both jaws, the molar region is most often affected [8–12]. Some authors have suggested that there is a female predilection [7,10–12], whereas others have reported no sex predilection [14,15].

The neoplasm is thought to be derived from the mesenchyme of a developing tooth or the periodontal ligament [8,11,16]. The odontogenic origin of OM is supported by its histologic similarity to the pulpal ectomesenchyme, proximity to the tooth-bearing areas of the jaws, periodic association with missing or impacted teeth, presence of inactive odontogenic epithelium in a minority of cases, and its uncommon occurrence in other parts of the skeleton [17].

OM is locally aggressive and invasive with the potential for persistent local growth and bone destruction. The tumoral potential to destroy bone extensively also includes its ability to invade the adjacent soft tissues. The standard treatment is surgical removal with safe margins. Because of its infiltrative nature, OM is prone to recurrence if resection is inadequate. There is a high rate of local recurrence after curettage alone.

We present an unusual case of OM as a rapidly expanding lesion in the mandible of a 62-year-old woman who underwent segmental mandibulectomy and mandibular reconstruction using a titanium plate.

## 2. Case report

A 62-year-old woman was referred to our department with the chief complaint of a painless left gradually enlarging mandibular swelling that she noticed 4 months before the initial presentation. A rapidly growing swelling was found after extraction of a mobile left mandibular canine and first premolar by her dentist 2 weeks previously. Marked overall swelling of the left mandibular gingiva was seen intraorally (Fig. 1).

A panoramic radiograph showed a well-circumscribed, multilocular, elliptical radiolucency extending from the mandibular right central incisor to the left mandibular body region (Fig. 2). All teeth of the mandible were vital. The lesion involving the arborescent radiopaque content extended to the inferior border of the left

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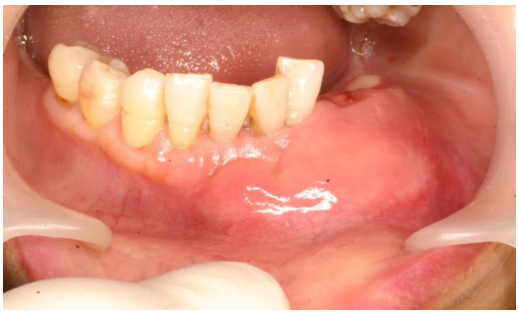

**Fig. 1.** An intraoral view shows the marked overall swelling of the left mandibular gingiva.

mandible. A computed tomography (CT) scan showed the soap-bubble appearance of the septated lesion and large extension of the buccal alveolar bone (Fig. 3). The lesion was about 70 mm × 35 mm in diameter. The buccal cortical plate was very thin, with no complete resorption. T1-weighted magnetic resonance imaging (MRI) showed a homogeneous low signal intensity (Fig. 4). T2-weighted MRI showed an inhomogeneous mass with high and low signal intensities.

A biopsy of the lesion was performed under the diagnosis of an odontogenic tumor and the histopathology confirmed the presence of OM of the mandible. The patient was treated for benign but locally aggressive mandibular pathology. Surgical treatment was segmental mandibulectomy with immediate plate reconstruction using a combined intraoral and extraoral approach. Under general anesthesia, a segmental mandibulectomy was performed through a combined intraoral and extraoral approach from the distal aspect of the right mandibular canine to the left mandibular angle with clear margins of about 15 mm on each side of the lesion (Fig. 5). The mandible was reconstructed with a 2.8-mm titanium plate immediately after the resection. The length of the resection was reflected in the length of the reconstruction plate. The reconstruction plate was bended during surgery, and the three terminal screw holes at each end served to fix the plate to the mandible remainder.

The lesion, which was in the mandible with no perforation of the buccal cortical plate, was solid and non-encapsulated. Microscopy showed stellate and spindle-shaped cells sparsely arranged in abundant mucoid stroma (Fig. 6). Only a few collagen fiber bundles were found in the mucoid-rich stroma. The intercellular material was positive with Alcian blue staining. Neither odontogenic epithelium nor calcified material was identified. Immunohistochemical staining was positive for vimentin and negative for desmin and p53, and Ki-67 labeling index was <5%. The current case had low expression of smooth muscle actin (α-SMA). OM was diagnosed based on the histologic findings. The patient was followed up for 24 months without a recurrence.

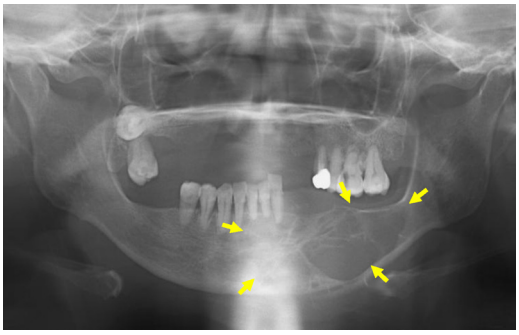

**Fig. 2.** A well-circumscribed, multilocular, elliptical radiolucency extending from the mandibular right central incisor to the left mandibular body region is seen in the panoramic radiographic image.

## 3. Discussion

OM is considered to be a rare, slow-growing, non-metastasizing tumor characterized by asymptomatic expansion of the jaw. The lesion is benign, but it is locally aggressive causing bone perforation, root resorption, tooth displacement, and mobility. Unusual cases of rapidly expanding OM of the jaw have been reported [18–20]. The rapid growth is believed to be due to production of a mucoid ground substance by stellate tumor cells [19,21]. In the current case, the tumor had been growing gradually for 4 months and appeared to grow rapidly after removal of the neighboring mobile teeth. It was suspected that the rapid expansion of the lesion was induced by the removal of mobile teeth.

It is generally accepted that OM develops in patients aged 10–50 years [2,3,7,9,11,12]. Most authors have agreed that there is a slight predilection for the posterior mandible, regarding the site of the tumor within the jaws [6–13]. The current case in a 62-year-old patient is extremely rare but typical of OM regarding location and sex. It was an unusual case of rapidly expanding OM following the extraction of the teeth in a 62-year-old patient.

Radiographically, OMs frequently appear as unilocular or multilocular radiolucencies with well-defined margins and fine bony septa. The lesion sizes are correlated with their locularity [22,23]. Lesions >40 mm tend to be multiloculated, and smaller lesions tend to be uniloculated [12,23]. The presentation often is described as a honeycomb, soap-bubble, tennis-racket, or ground-glass pattern. One report described that the tennis-racket appearance was exhibited mostly by the trabeculation of the multilocular lesions [12]. The current case had a soap-bubble appearance with interlaced bone trabeculae. The radiographic differential diagnosis of multilocular OM should include ameloblastoma, central giant cell granuloma, intraosseous hemangioma, aneurysmal bone cyst, cherubism, osteosarcoma, metastatic neoplasm of the jaws [22], keratocystic odontogenic tumor, and glandular odontogenic cyst [24]. The clinical differential diagnosis of OM included ameloblastoma due to the feature of rapid expansion and age of the patient. The current case of OM was difficult to distinguish from the more common forms of odontogenic lesions. The diagnosis of OM was reached by histopathologic examination of the biopsy specimen.

Histologically, OM is a hypocellular tumor comprising stellate and spindle-shaped cells lying loosely in abundant myxoid stroma. In the current case, myxoid stroma also contained a few collagen fibers, with no calcified materials or odontogenic epithelial nests. No histological differences between the central portion and the peripheral expanding front portion were found.

To establish a precise diagnosis, immunohistochemical staining is most reliable for differentiating OM from other lesions including malignant tumors, such as myxofibrosarcoma. The immunohistochemical reaction of the tumor cells to antibodies varies [10,25–27]. In most cases of OM of the jaw, the tumoral cells usually express SMA [10]. The current stained positively for SMA. Vimentin as a fibroblastic marker was positive, and desmin and p53 were negative. Desmin was stained for the confirmation of differentiation of spindle-shaped cells. Ki-67 antigen, found in proliferating cells, identifies the growth fraction of normal and tumor cells, and it can be utilized as an indicator of biological behavior of disease and an important biomarker related to prognosis. The Ki-67 labeling index was <5%. The immunohistochemical findings were useful for diagnosing OM.

The recommended treatment for OM depends on the lesion size and behavior and can vary from enucleation or curettage to radical resection [8,10,28]. Due to its locally invasive nature, OM of the jaws tends to be treated by bone resection including peripheral ostectomy and segmental mandibulectomy [20,29]. These techniques remove a circumferential margin of bone around