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## Case Report

# Odontogenic myxoma: A dual affliction in maxilla and mandible - A case report

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## ABSTRACT

The odontogenic myxoma (OM) is a locally invasive benign odontogenic tumour of the jaws, thought to originate from the mesenchymal portion of the tooth germ. It commonly manifests in individuals during their second or third decade of life.

Females experience a higher frequency of occurrences compared to males, and there is a greater inclination for odontogenic myxoma to affect the mandible rather than the maxilla. In most cases, OM manifests as an asymptomatic swelling. Due to its non-capsulated and aggressive nature, there is a notable high recurrence rate reported.

Small myxomas are typically addressed through curettage, but it is crucial to conduct thorough follow-up examinations for a minimum of 5 years. The standard treatment for myxoma typically involves surgical resection.

Our article highlights two instances of odontogenic myxoma occurring in separate locations. The emphasis is on summarizing distinctive clinical and radiological behaviour of these lesions manifesting in the maxilla and mandible.

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## 1. Introduction

The odontogenic myxoma (OM), a benign non-encapsulated odontogenic tumor, was first described by Thomas and Goldman in 1947.<sup>1</sup> German pathologist Rudolph Virchow coined the term "myxoma" in 1871 to characterise tumors that shared similar histological characteristics with the mucinous tissue of the umbilical cord.<sup>1</sup>

According to the 2022 World Health Organization Classification of Odontogenic Lesions, OM comes under benign mesenchymal odontogenic tumor.<sup>2</sup> It is a rare tumor, with or without the presence of odontogenic epithelium, and is referred to as "locally malignant" due to its remarkably high local aggressiveness and non-metastasizing nature,<sup>3,4</sup> with a low prevalence of 3-6% of odontogenic tumor.<sup>1,3</sup>

Radiographically, OM appears as a well-defined unilocular or multilocular osteolytic lesion with the appearance of a "soap bubble".<sup>5,6</sup> The distinguishing feature associated with this tumor is the presence of a straight, thin, finely detailed septum. While these septa have been likened to a tennis racket or stepladder pattern, such configurations are infrequently observed. In reality, the majority of septa exhibit a curved and coarse structure. However, the identification of one or two of these straight septa significantly aids in recognizing this tumor.<sup>7</sup>

## 2. Case Report

### 2.1. Case 1

A 40-year-old male reported to our department with pain and swelling on the right mandible, following a bike fall a year ago. Initial trauma led to a pea-sized swelling,

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progressively enlarging over a year, causing noticeable facial asymmetry. Intermittent pain and pus discharge were observed. Upon extraoral examination, a diffuse ovoid swelling of size approximately 8\*7 cm was present on the right lower half of the face that extended supero-inferiorly from the ala-tragus line to the lower border of the mandible and antero-posteriorly from the right corner of the mouth to the angle of the mandible (Figure 1). The swelling bony hard & tender on palpation showed local warmth, ipsilateral submandibular lymph node enlargement. Upon intraoral examination vestibular obliteration in relation to 46, 47, and 48 was seen (Figure 2). Routine hematologic tests were normal. Radiographic assessments (OPG, CECT, CBCT) and a biopsy were performed for histopathological diagnosis.



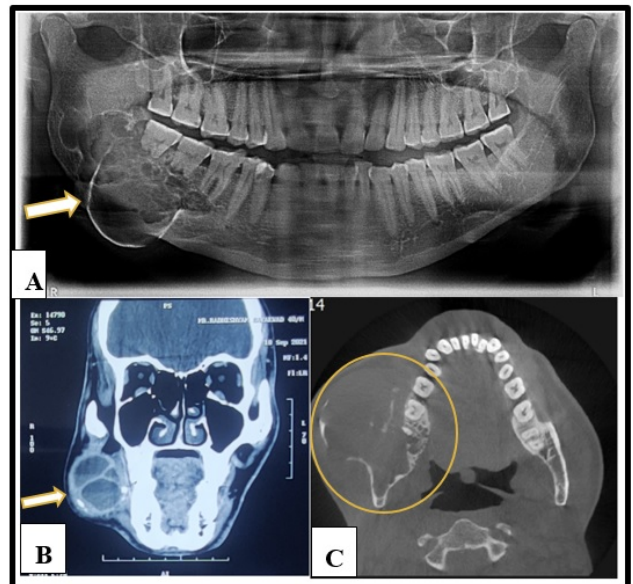
**Figure 2:** Intraoral image of patient 1 showing vestibular obliteration in relation to 46, 47, and 48



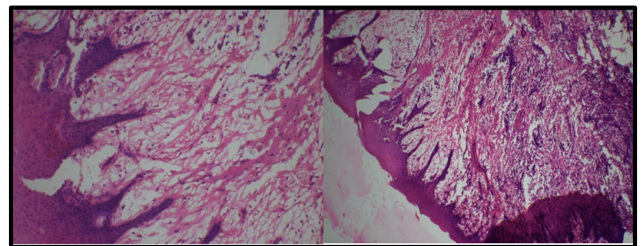
**Figure 1:** Extraoral image of patient 1 showing a diffuse ovoid swelling of size approximately 8\*7 cm present on the right lower half of the face extending supero-inferiorly from the ala-tragus line to the lower border of the mandible and antero-posteriorly from the right corner of the mouth to the angle of the mandible

Radiograph revealed significant destruction in the mandibular body, characterized by a well-defined, predominantly radiolucent lesion, with multiple septations in the right mandibular body, angle and ramus region giving soap bubble appearance. Root resorption was evident in relation to teeth 46, 47, and 48. Buccal cortical plate is expanded and thinned, and perforated, blends in the adjacent soft tissue. Lingual cortical plate thinned in 46, 47, 48 regions (Figure 3).

Fine Needle Aspiration Cytology (FNAC) and biopsy confirmed a histopathological diagnosis of odontogenic myxoma, characterized by spindle-shaped mesenchymal cells in a myxoid stroma (Figure 4).



**Figure 3:** Radiographic image of patient 1 (A) OPG (B) CT PNS (C) CBCT axial section showing a well-defined, predominantly radiolucent lesion, with multiple septations in right mandibular body region extending mesiodistally from distal root surface of 46 till the posterior ramus region; root resorption evident in relation to 46, 47,48



**Figure 4:** Histopathologic image of patient 1 showing spindle-shaped mesenchymal cells in a myxoid stroma

## 2.2. Case 2

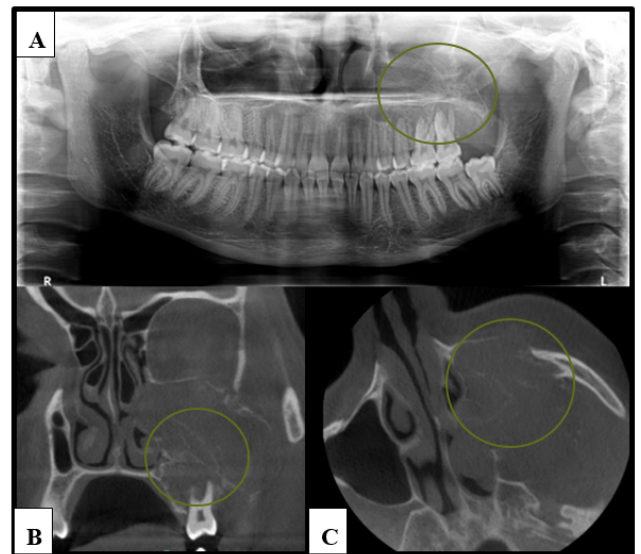
A 19-year-old female reported to our department with persistent left facial swelling for one year. One year ago, the same swelling was diagnosed as pyogenic granuloma, and the extraction of tooth 28 was performed at that time. Despite the procedure, the swelling persisted, leading to mild facial disfigurement with time prompting the patient to seek our evaluation. Upon extraoral examination, a diffuse swelling of size of approximately 5\*5 cm was present on the left malar region that extended superoinferiorly from the infraorbital margin to the angle of the mandible and anteroposteriorly from the left corner of the mouth to the anterior surface of the ramus of the mandible (Figure 5). The swelling was tender to palpate and bony hard in consistency. Intraoral examination revealed a single well-defined swelling present on the 28 region up to the retromolar pad region of size approximately 1\*1cm, and vestibular obliteration from the 24 up to the 27 region (Figure 6). No tooth displacement or mobility observed. Routine blood tests were normal. Diagnostic assessment included OPG, PNS imaging, CBCT, and biopsy. Radiographic analysis revealed multilocular irregular radiolucent lesion in left posterior maxilla with internal trabecular pattern which was changed to thinning of trabeculae and widening of marrow spaces giving appearance of honeycomb pattern. Buccal and palatal cortical plates were thin and expanded. Thinning & destruction observed with floor and posterior wall of the left maxillary sinus, inferior orbital rim in left side & left lateral wall of nose was observed. No root resorption was observed (Figure 7). Histopathological findings were indicative of odontogenic myxoma.



**Figure 5:** Extraoral image of patient 2 showing a diffuses welling of size approximately 5\*5 cm present on the left malar region extending superoinferiorly from the infraorbital margin to the angle of the mandible and anteroposteriorly from the left corner of the mouth to the anterior surface of the ramus of the mandible



**Figure 6:** Intraoral image of patient 2 showing a single well-defined swelling present in 28 region up to retromolar pad region of size approximately 1\*1cm, and vestibular obliteration from 24 to 27 region



**Figure 7:** Radiographic image of patient 2 (A) OPG (B) CBCT coronal section (C) CBCT axial section showing multilocular, irregular, radiolucency in left posterior maxilla; involving 25-27, maxillary tuberosity & pterygoid plate region with appearance of honeycomb pattern. Thinning with breach is observed with inferior orbital rim in left side

## 3. Discussion

The OM is an uncommon benign tumour that grows slowly and has little chance of developing into something malignant, believed to originate from the ectomesenchyme of a developing tooth germ due to its resemblance to the dental follicle and papilla.<sup>8</sup>

Odontogenic myxoma (OM) typically manifests between the second and fourth decades, with an average diagnosis age of 29.6 years according to Kaffe et al.<sup>1</sup> OM is more common in women (1:2 ratio), with a mandible-to-maxilla ratio of 4/3.<sup>1</sup> It most commonly manifests in the posterior mandible. OM often presents asymptotically as a painless swelling, reaching a



considerable size before patient awareness prompts treatment. Uncommon symptoms in advanced cases include pain, tooth displacement, and paresthesia.<sup>4</sup> Mandibular OM frequently results in bony expansion that causes teeth to become loose, root resorption, malocclusion, poorly fitting dentures, and facial deformity.<sup>1</sup> Invasion of the antrum occurs frequently in lesions of the maxilla.

Radiographically, OM appears as a well-defined unilocular or multilocular osteolytic lesion with the appearance of a "soap bubble" or "honeycomb" or "tennis racquets" with expansion of cortical bone and displacement of the associated tooth can appear in a long-standing OM.<sup>5,6</sup> Fine trabeculations within small lobules are typically visible in the honey-comb type of myxoma, but they are absent in ameloblastoma.<sup>1</sup>

Myxoma's aetiology is unknown. It is a typically slow-growing neoplasms; nevertheless, it has been demonstrated that local trauma may accelerate their growth.<sup>1</sup> Histological examination serves as a diagnostic tool for myxoma.<sup>4</sup>

Optimal treatment for odontogenic myxoma (OM) is debated. Major surgery, such as segmental resection, is favored for local aggressiveness and potential bone destruction. However, comprehensive approaches like excision with curettage alone may be inadequate due to OM's non-encapsulated nature and infiltration of surrounding bone. Because of the significant risk of recurrence, which is said to range from 10% to 33%,<sup>8</sup> surgical resection is the only initial treatment option for prolonged OM.<sup>9</sup> Odontogenic myxomas (OM) show PRKAR1A gene mutations and reduced protein expression. Identifying pathogenic genes may enable molecular treatments, preventing deformities resulting from surgery.<sup>10</sup>

#### 4. Conclusion

Odontogenic myxoma (OM), a rare, slow-growing benign jaw tumor, exhibits aggressive behavior. Early detection and prompt treatment are crucial to prevent extensive site destruction. OM can be diagnosed on the basis of clinical and radiographic analysis but confirmed on the basis of histopathological reports. So OM requires a multidisciplinary approach for the diagnosis and treatment.

#### 5. Source of Funding

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#### 6. Conflict of Interest

None.


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