

# Odontogenic Myxoma with Displaced Third Molar to Coronoid Process: Role of Differential Stains to Assess Biologic Behavior

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## ABSTRACT

**Aim:** The aim of this case report is to document an aggressive odontogenic myxoma (OM) of the mandible causing dramatic displacement of the unerupted third molar up to the coronoid process, a rare entity.

**Background:** The OM is an aggressive neoplasm that can cause significant destruction of the jaws. They present as large multilocular lesions often causing thinning of the cortical plates and displacement of teeth. The OM has a high rate of recurrence that brings into significance their mode of management that varies from curettage to radical excision depending on the surgeon's school of thought.

**Case description:** An OM of the mandible in a 17-year-old patient is described here with cone-beam computed tomography (CBCT) and orthopantomograph (OPG) findings. The OM had displaced the unerupted mandibular third molar to the coronoid process that makes this case interesting to report.

**Conclusion:** Intensity of alcian blue and safranin O staining reveal the hyaluronic acid, glycosaminoglycans (GAG's), and proteoglycan content of OM. The pattern of picosirius red staining under polarizing microscope indicates aggressive potential of OM.

**Clinical significance:** The article highlights the use of appropriate differential stains like alcian blue, safranin O, and picosirius red that facilitate in arriving at precise biologic behavior of OM. Original research studies to validate intensity and pattern of special stains with odontogenic lesions are mandated to establish a significant correlation.

**Keywords:** Alcian blue, Cone-beam computed tomography, Odontogenic myxoma, Picosirius red, Polarizing microscope, SafraninO, Tooth displacement.

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## INTRODUCTION

Odontogenic tumors represent a broad spectrum of lesions ranging from benign to malignant. Benign odontogenic lesions are rare entities that are important due to their local aggressive nature and equally challenging to handle. Odontogenic myxoma (OM) of the jaws was first described by Goldman and Thoma in 1947. It represents 3 to 6% of all odontogenic tumors.<sup>1-4</sup>

Odontogenic myxomas are believed to originate from the mesenchymal portion of the odontogenic apparatus. In earlier days few researchers believed that the OM and osteogenic myxoma were two separate entities. However, with evidence-based research, it is clear that all myxomas of jaws are of odontogenic origin.<sup>5</sup>

The OM is an aggressive lesion that frequently expands the bone and causes destruction of the cortical plates, it is often extensive in involvement. Displacement of teeth by OM is a relatively common finding. Hodson and Prout have suggested that the presence of large amounts of hyaluronic acid is a significant factor for the aggressive biologic behavior of the lesion.<sup>6</sup>

Odontogenic myxoma is infamous for a high recurrence rate of up to 25% after curettage.<sup>6</sup> A minimum follow-up period of 5 years without recurrence is recommended by some researchers before performing reconstructive surgeries. Special stains have been seldom used to demonstrate the aggressive potential of the lesion. The aim of this article is to report a large OM with a unique displacement of the unerupted mandibular third molar to the coronoid process. The report emphasizes the role of differential stains that relate to the biologic behavior of the lesion.

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## CASE DESCRIPTION

### Clinical Presentation

A 17-year-old male patient visited the dental clinic with a chief complaint of swelling on the right side of the jaw for 2 months. A diffuse ill-defined swelling was seen on the right side of the mandible extending from the symphysis to the body of the right mandible causing facial asymmetry (Figs 1A and B). The swelling was hard in consistency and slightly tender. On intraoral examination, a solitary large swelling was seen extending from

the 42-tooth region to the distal root of 47 obliterating the buccal vestibule (Fig. 1C). It was firm in consistency, immobile with overlying intact mucosa.



**Figs 1A to D:** (A) A diffuse ill-defined swelling was seen on the right side of the mandible; (B) Swelling causing facial symmetry; (C) Solitary large swelling was seen extending from the 42-tooth region to the distal root of 47 obliterating the buccal vestibule; (D) Incisional biopsy from the lesion

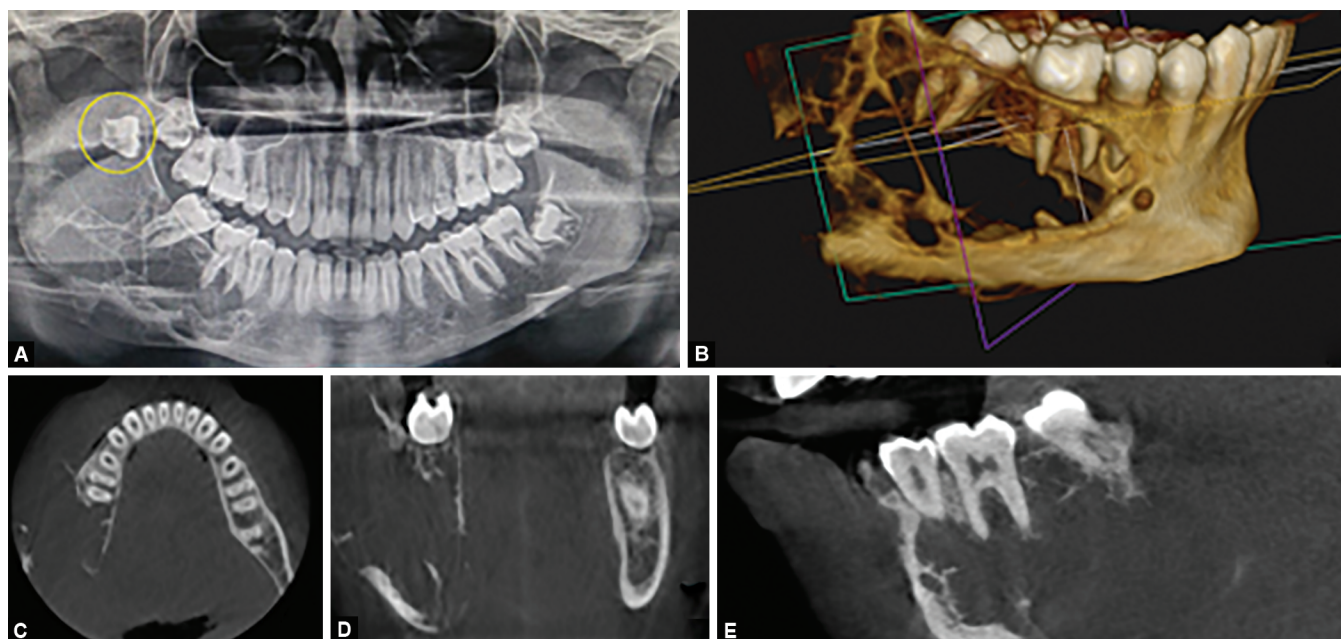
## Radiographic Investigations

A panoramic radiograph was taken which revealed a large well-defined multilocular lesion involving the right body and ramus of the mandible. The radiolucency measured approximately  $6 \times 4$  cm and extended anterior posteriorly from the distal aspect of tooth number 45 along the posterior border of ramus up to the coronoid process and superior inferiorly extended from the coronoid process to the inferior border of mandible. The radiolucency showed displacement of the impacted mandibular third molar to the coronoid process (Fig. 2A). The internal structure of the lesion revealed angular and multilocular compartments separated by straight septa forming triangular, square, and rectangular spaces.

To reveal the complete extent of the lesion, the patient was scanned using Carestream (CS3D) machine at 90 kVp, 8 mA with an exposure time of 8 seconds and  $10 \times 10$  cm diameter scan volume. A 3D image of the lesion revealed buccal cortical plate perforation and internal septae (Fig. 2B). Axial sections of the cone-beam computed tomography (CBCT) showed buccal cortical plate perforation (Fig. 2C). Coronal sections of the CBCT images showed buccal cortical plate expansion with sharp septa (Fig. 2D). Sagittal sections of the CBCT revealed the multilocular nature of the lesion with internal septae along the periphery of the lesion (Fig. 2E). These findings prompted a radiographic diagnosis of OM for the lesion.

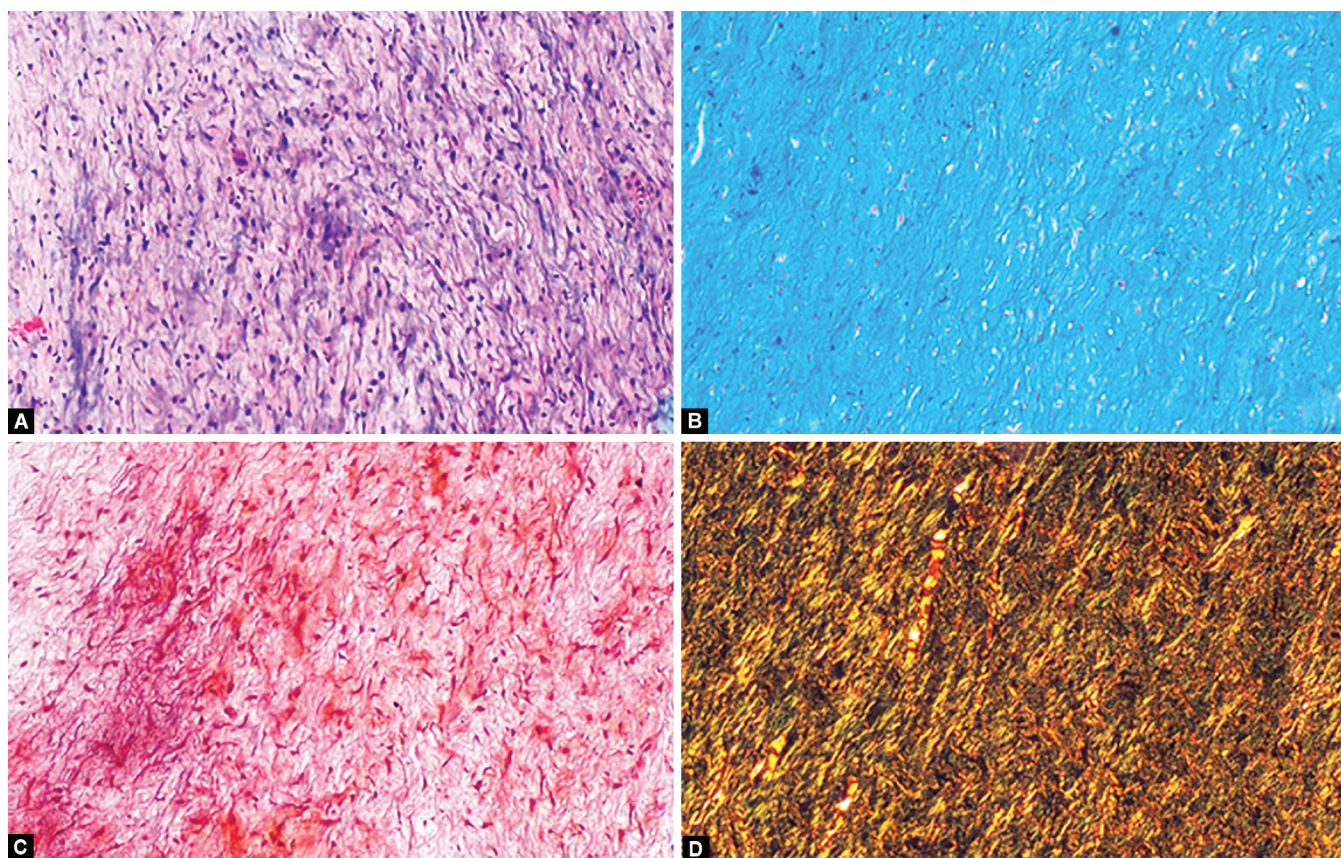
## Histopathology

An incisional biopsy was performed with aseptic precautions (Fig. 1D). The tissue obtained was glistening and jelly like. On histopathological examination, stellate and spindle-shaped cells were observed in an abundant, loose myxoid stroma (Fig. 3A). Odontogenic epithelial islands were seen. A final diagnosis of OM was established. The following special stains—Alcian blue, safranin O, and picrosirius red—were employed. A high intensity



**Figs 2A to E:** (A) Panoramic radiograph reveals a large well-defined multilocular radiolucency exhibiting a tennis racket appearance with displacement of the impacted mandibular third molar to the coronoid process; (B) 3D image of the lesion revealing the buccal cortical plate perforation and internal septae; (C) Axial sections of cone-beam computed tomography (CBCT) show buccal cortical plate perforation; (D) Coronal Sections of the CBCT images show buccal cortical plate expansion with sharp septa; (E) Sagittal sections of the CBCT revealing the multilocular nature of the lesion with internal septae along the periphery of the lesion





**Figs 3A to D:** Photomicrographs of odontogenic myxoma: (A) Section showing stellate and spindle-shaped cells in an abundant loose myxoid stroma with basophilic hue (H&E stain,  $\times 100$ ); (B) Intense staining of the loose myxoid stroma demonstrating hyaluronic acid and glycosaminoglycans (Alcian Blue stain,  $\times 100$ ); (C) Intense staining of the myxoid stroma indicating proteoglycan content (Safranin O stain,  $\times 100$ ); (D) Section showing greenish yellow birefringence under a polarizing microscope (Picrosirius Red stain,  $\times 100$ )

of staining was observed with Alcian blue (Fig. 3B) and safranin O (Fig. 3C). Picrosirius red also demonstrated intense staining, and the same was observed under a polarizing microscope that exhibited a greenish yellow birefringence (Fig. 3D). The staining intensity of Alcian blue and safranin O with the polarizing pattern of picrosirius red correlated with the aggressive potential of the lesion. Taking into consideration the size of the lesion and its potential for recurrence, a segmental resection has been planned with reconstruction.

## DISCUSSION

Odontogenic myxomas are aggressive odontogenic tumors that are usually found in the second to third decade of life. Odontogenic myxoma has a mandibular predilection with a rapid clinical growth, smaller lesions however may be asymptomatic and discovered on routine radiographs. Its infiltrative growth pattern and aggressive growth potential with high recurrence rate makes it a significant lesion to reckon with.<sup>7</sup> The larger aggressive lesions are usually associated with large amounts of myxoid ground substance in the tumor matrix.

The OM's can grow to a considerably significant size, few researchers have reported lesions over 6 cm in size. A case report by Liu et al. described a lesion measuring approximately  $16 \times 16$  cm involving the maxilla bilaterally.<sup>8</sup> A study by Simon et al. analyzed 33 cases of OM's in a Tanzanian population and concluded that

magnitude of the symptoms were in accordance with the size of the tumor.<sup>9</sup> In the present case, the tumor measured  $6 \times 4$  cm causing asymmetry of the face.

Displacement of teeth by few millimeters by the tumor mass is a relatively common finding, but root resorption is less frequent. Minor displacement of teeth has been registered in 9.5% of the cases.<sup>10</sup> According to some studies, root displacement rather than resorption is the rule of jaw myxomas.<sup>11–13</sup> This is supported by the results of the by Li et al., where tooth/root displacement was noted in 11 (47.8%) of cases but root resorption in only 3 (13%).<sup>14</sup> The present case reports a dramatic displacement of the unerupted mandibular third molar upto the coronoid process. This is a highlight of the present report, as no OM case in the literature has reported till date as the displacement of an unerupted mandibular third molar to the coronoid process till date.

Radiographically, most myxomas appear as a multilocular lesion with separation by thin bony septae to give a tennis racket or honey comb appearance. Zang et al. have classified radiographic appearances of OM's into six types—Type I: Unilocular, Type II: Multilocular (honey comb, soap bubble or tennis racquet pattern), Type III: Lesion located in alveolar bone, Type IV: Lesion involving the maxillary sinus, Type V: osteolytic destruction, and Type VI: Combination of bone destruction and bone formation.<sup>15</sup> Kaffe et al. in his radiographic study revealed correlation between size and locularity, unilocular lesions were smaller than 4 cm, and multilocular lesions were larger than 4

cm.<sup>16,17</sup> The present case exhibited osteolytic destruction (Type V) with displacement of the impacted mandibular third molar to the coronoid process.

However, plain radiography is not reliable enough to demonstrate the extent and fine inner structures of the tumor.<sup>18</sup> CBCT is highly effective in demonstrating the comprehensive internal structures of the lesions precisely. As reported by Wang et al., CBCT reveals a mixed radiolucent–radiopaque internal pattern of the tumor, and it is considered as the characteristic feature of OM. The authors examined CBCT reports of 18 patients and stated that fine and straight septa were recognized that separated the tumor into triangular, square, or rectangular spaces. CBCT was effective in demonstrating the inner structure of OM.<sup>19</sup> Owing to the high rate of recurrence, the accurate radiographic appearance was critically important in arriving at a correct diagnosis and in surgical planning. In the present case, CBCT revealed the multilocular nature of the lesion with internal septae along the periphery of the lesion which indicated toward the diagnosis of OM.

The differential diagnosis of OM includes ameloblastoma, central giant cell granuloma, central hemangioma, osteosarcoma, and on few occasions odontogenic keratocyst. Microscopic examination leads to a conclusive diagnosis, as the histopathologic features of OM are characteristic.

The OM is infiltrative with no capsulation. They are made up of loosely arranged, spindle-shaped, and stellate cells with long intermeshing fibrillar processes. The intercellular substance is usually mucoid. Multiple tiny capillaries are usually found. Nests or islands of odontogenic epithelium may be observed infrequently.<sup>20</sup> Similar features were observed in the current case.

Special stains have been performed in few OM cases reported in the literature. Alcian blue stains GAG's and hyaluronic acid in the stroma (ground substance) of OM. Safranin O stains the proteoglycans present in OM. These materials when present in excess are responsible for the aggressive biologic behavior of myxomas.<sup>21,22</sup>

The staining intensity of Alcian blue and safranin O in the present case indicated a high content of GAG's, hyaluronic acid, and proteoglycans. Picrosirius red revealed intense staining and greenish–yellow birefringence under polarizing microscope. The change in birefringence from red to greenish yellow specifies an aggressive potential of the lesion.<sup>23</sup> The intensity (alcian blue and safranin O) and pattern of special staining (Picrosirius red-polarizing microscope) correlated well with the aggressive potential of the current case which had increased to a considerable size.

Recommended therapy varies from curettage to radical excision. Complete surgical removal can be difficult. Boffano et al. suggested that lesions of size >3 cm are considered for radical resections and bloc resections and tumors of less diameter are better treated by enucleation or curettage.<sup>24</sup> These characteristics may explain the high rate of recurrence of myxomas, which ranges from 10% to 33% with an average of 25%.<sup>14,15</sup>

Francisco et al. conducted a clinicopathologic analysis of 14 cases of OM and concluded that there is no gold standard surgical management, and the therapeutic decision should be individualized, taking into account the characteristics and extension of the tumor.<sup>25</sup> In the present case, a segmental resection has been planned.

Although the OM is a benign lesion, its treatment should be aggressive due to its high recurrence mainly due to its penetrative

growth pattern. Follow-up of cases is mandatory. Owing to its indefinite behavior coupled with diagnostic and management dilemmas related to myxomatous lesions, a sound knowledge with accurate histopathological diagnosis is required for effective management of OM's.

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