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Case Report Article

Odontogenic myxoma in the maxilla: clinical case report

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Abstract

Introduction: Odontogenic myxoma is a rare, benign tumour of ectomesenchymal origin, characterised by locally invasive behaviour, with treatment directly related to recurrence. This neoplasm is uncommon and grows slowly. Despite its benign nature, its characteristics can lead to facial deformities and significantly impact patients' quality of life. **Objective:** This study reports a case of odontogenic myxoma in the maxilla of a 15-year-old male. **Case report:** The patient who attended a stomatology clinic complaining of asymptomatic swelling of the left cheek. On clinical examination, an increased volume was observed below the left orbit, along with orbital asymmetry and an intraoral tumour. Complementary exams, including radiography, tomography, vitality testing, and needle aspiration, suggested odontogenic myxoma, later confirmed by incisional biopsy and histopathological examination. The patient was referred for surgical resection and remains recurrence-free after 20 months, having completed rehabilitation. **Discussion:** This case highlights the importance of precise diagnosis and an individualised treatment plan to optimise prognosis and improve patients' quality of life. **Conclusion:** Odontogenic myxoma requires careful diagnostic and therapeutic approaches due to its locally invasive behaviour and potential impact on patients' function and aesthetics.

Introduction

Odontogenic myxoma (OM) is an uncommon, slow-growing mesenchymal or ectomesenchymal odontogenic tumour [15]. While its histogenesis remains uncertain, it is likely associated with the odontogenic ectomesenchyme of a developing tooth or the periodontal membrane [3, 7, 15]. This lesion is known to exhibit infiltrative biological behaviour, yet it is incapable of metastasizing to other regions [10].

OM is more prevalent in females, with a ratio of 1:36, typically occurring in the second to fourth decades of life, and predominantly affecting the posterior region of the mandible [1, 4, 10]. Clinically, OM is characterised as a slow-growing, asymptomatic lesion with progressive expansion [1, 15]. In advanced stages, swelling in the affected region may be observed, potentially leading to pain, root resorption, facial asymmetry, mandibular nerve paraesthesia, malocclusion, tooth mobility, unerupted teeth and masticatory disturbances [6]. Additionally, expansion into the maxillary sinus, as well as involvement of the palate, orbit and nasal cavity, may occur [10].

The diagnosis is established through clinical examination, imaging studies, and histopathological analysis. Radiographically, OM most frequently presents as a radiolucent image with poorly defined borders, a multilocular appearance and fine trabeculations [4, 5]. However, unilocular radiolucent images, usually of smaller dimensions, may also be observed [4]. Macroscopically, the lesion exhibits a mucoid or gelatinous structure [8, 13]. Microscopically, the tumour is composed of stellate, spindle-shaped and rounded cells that are irregularly distributed within an abundant, loose and fibrillar myxoid or mucoid stroma with sparse collagen fibrils. Occasionally, small islands of inactive odontogenic epithelial remnants may be present within the myxoid substance, although their identification is not essential for diagnosis [9].

The treatment of OM is not well-established, and its management remains challenging. Therapeutic approaches range from simple curettage, resection, curettage with Carnoy's solution or enucleation with curettage, to surgical excision [11].

Although benign, OM exhibits aggressive potential and may recur due to its lack of encapsulation and loose consistency, despite generally having a favourable prognosis [2, 11].

Given that OM is a rare benign mesenchymal tumour that can involve facial hard tissues, presents with nonspecific features and may resemble other intraosseous lesions, it can lead to delayed diagnosis and has a high recurrence rate. These factors significantly impact treatment choices and patient prognosis. Case reports, such as the present study, are essential to expand knowledge and improve clinical practices for dental surgeons across all specialties. The aim of this study is therefore to report the case of a 15-year-old patient diagnosed with odontogenic myxoma in the posterior maxilla and detail its clinical, imaging and histopathological features, as well as the surgical and rehabilitative management, with a 20-month follow-up period.

Case report

A 15-year-old male presented to the stomatology clinic, complaining of "swelling in the cheek". During the anamnesis, he reported no pain, but noted the swelling began approximately eight months prior. No significant semiological alterations, habits or vices were reported. An extraoral physical examination revealed left facial asymmetry, atypical facies and swelling below the left orbital region that caused orbital displacement (figure 1A). An intraoral examination identified a sessile-based tumoral lesion with a colour similar to the mucosa, firm consistency and poorly defined borders. The lesion extended to the vestibule in the region of teeth 25 through 28, reaching the midline of the hard palate (figure 1B).

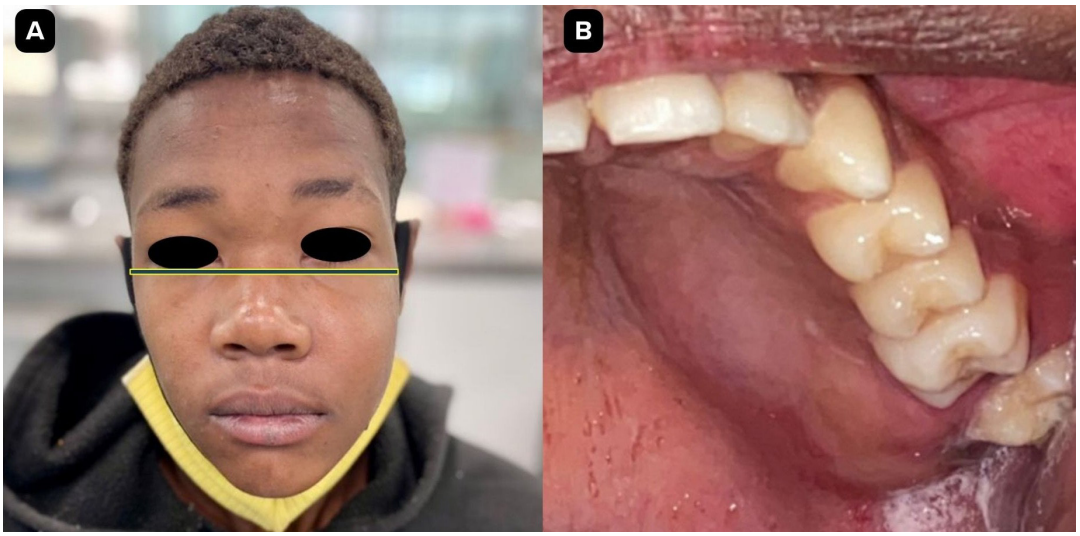

Figure 1 – A: Extraoral physical examination; B: intraoral physical examination

Following the clinical evaluation, a panoramic radiograph was requested (figure 2A). This revealed a unilocular radiolucent image with poorly defined borders, no radiopaque halo and intimate contact with the roots of teeth 23 through 28. Residual thin bone trabeculae were observed, arranged at right angles and resembled a tennis-racket pattern.

A cone-beam computed tomography (CBCT) scan was performed, revealing a voluminous hypodense, multilocular image that was partially circumscribed and partially delimited by a hyperdense halo (figure 2B). Numerous bone trabeculae were observed within the lesion, arranged at right angles in the left maxilla which involved the body and posterior region of the maxilla, including the root regions of teeth 23 through 27 and tooth 28. Discontinuity of the lamina dura of the involved teeth was noted. The lesion measured approximately 56.1 mm × 52.7 mm × 46.1 mm (i.e., superior-inferior, anteroposterior and medio-lateral dimensions, respectively), causing divergence of the roots of tooth 26 mesially and tooth 27 distally, extrusion of tooth 26 and displacement of the developing tooth 28 superiorly and posteriorly, adjacent to the lateral lamina of the pterygoid process of the sphenoid bone.

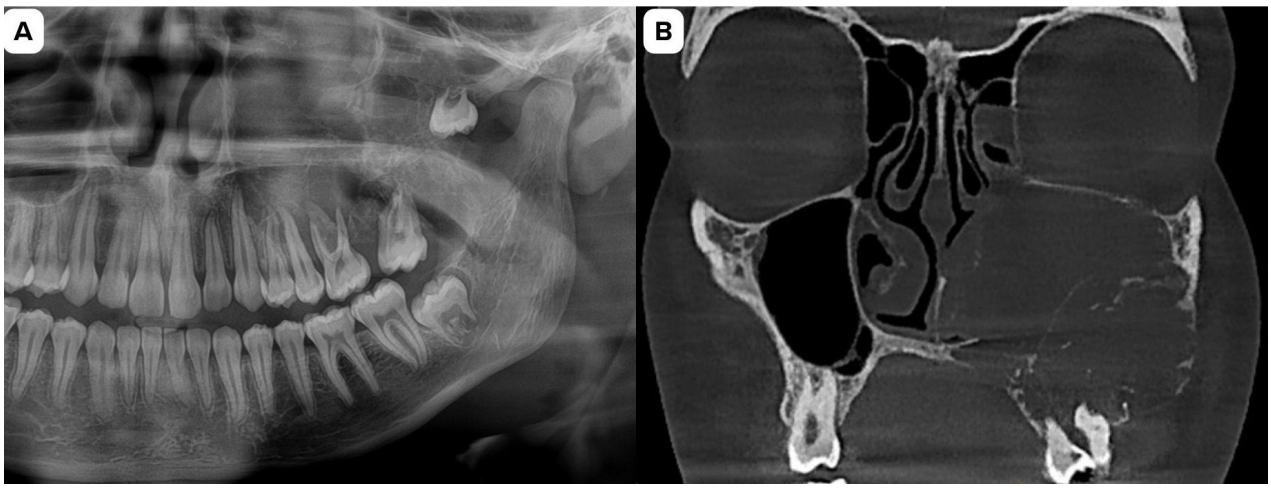

Figure 2 – A: Panoramic radiography; B: cone-beam computed tomography: coronal slice

Expansion and thinning of the buccal, palatal and alveolar cortices were observed, along with significant bulging and discontinuities of the left maxillary sinus floor, resulting in a bucco-sinus communication and sinus opacification due to fluid accumulation. Additionally, the lesion caused bulging and discontinuity of the lateral-basal wall of the left nasal cavity, involving the inferior and middle nasal conchae with obliteration of the corresponding meatus, bulging of ipsilateral ethmoid cells and bulging and discontinuity of the orbital floor.

Following the imaging evaluation, a pulp vitality test was performed due to the lesion's proximity to the apical regions of the affected teeth, which were found to be vital. Aspiration puncture was performed, yielding no fluid content. Based on the clinical findings, medical history and imaging results, the diagnostic hypothesis was odontogenic myxoma. An incisional biopsy was performed, and the specimen was sent for histopathological analysis.

Histological sections revealed a fragment of a mesenchymal odontogenic tumour characterized by the proliferation of randomly arranged stellate, spindle-shaped and ovoid cells within an abundant, loose stroma containing collagen fibrils and basophilic material consistent with extracellular matrix, which confirmed the diagnosis of odontogenic myxoma (figure 3).

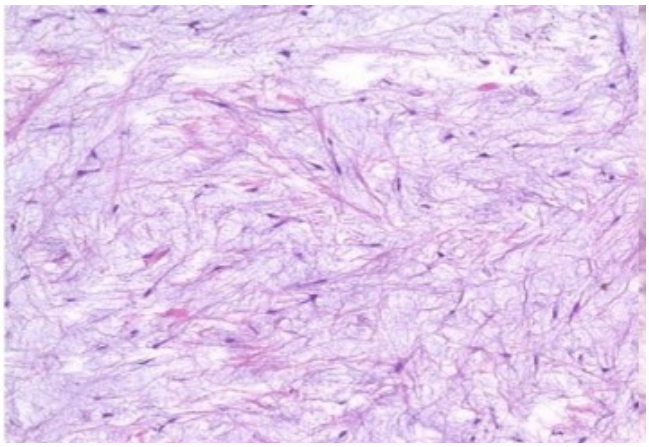

Figure 3 – Microscopic aspect of odontogenic myxoma (H&E X10 magnification)

The patient was referred to the Buccomaxillofacial Surgery Service, where he underwent surgical resection of the tumour under general anaesthesia. During the procedure, it was confirmed that the tumour did not involve the orbital floor, which allowed for a more conservative approach. The lesion was completely removed, albeit with reduced safety margins, considering the patient's age and potential future complications (figures 4A and 4B).

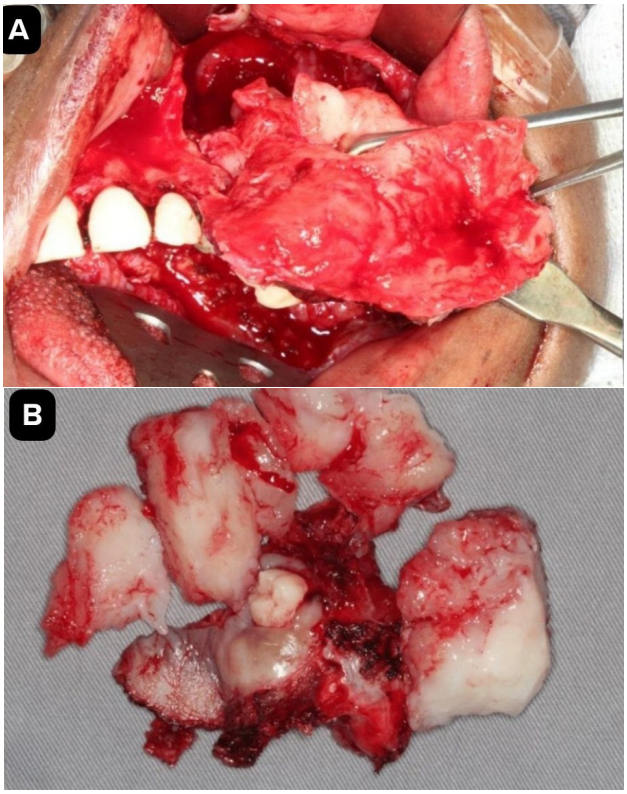

Figure 4 – A: Complete removal of the lesion; B: surgical specimen

One month post-surgery, the patient returned to the stomatology clinic. An extraoral examination revealed no facial alterations, with restored facial symmetry and orbital alignment. An intraoral examination showed good healing, although a small bucco-sinus communication persisted (figure 5A). During this evaluation, tooth 23 exhibited grade-3 mobility, and a panoramic radiograph confirmed bone loss, raising doubts about the tooth's prognosis in the rehabilitation process (figure 5B).

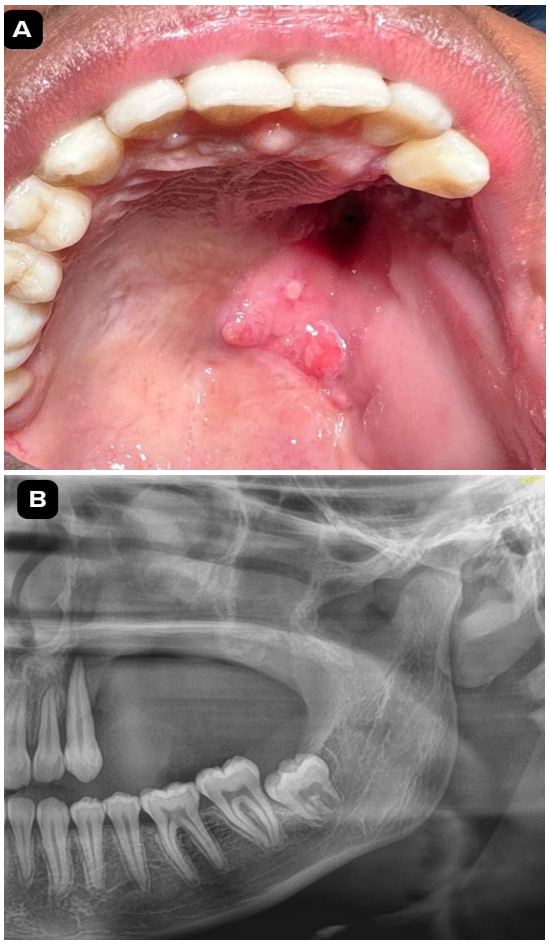

Figure 5 – A: 20-month clinical follow-up intraoral physical examination; B: 20-month radiographic follow-up panoramic radiography

A follow-up CBCT scan was requested, which confirmed complete tumour removal with a resultant bone defect and no evidence of recurrence. Notably, bone rarefaction involving tooth 23 was observed, which was consistent with local inflammatory/infectious processes. At the 20-month follow-up, complete closure of the bucco-sinus communication was observed, with satisfactory healing. A new panoramic radiograph confirmed no recurrence of the odontogenic myxoma.

For complete aesthetic and functional restoration, a customized removable partial denture (RPD) was fabricated following the extraction of tooth 23, using tooth 22 as a direct abutment (figure 6A).

Teeth 14 through 17 were used as indirect abutments to ensure greater stability. According to the universal classification, this is a unilateral posterior maxillary RPD with a tooth-mucosa-supported functional base, corresponding to Kennedy Class II.

Frontal view shows an adequate buccal corridor, with artificial teeth matching the patient's aesthetics, effectively restoring masticatory and phonetic functions and facilitating social reintegration (figure 6B).

The patient remains under follow-up with a good prognosis.

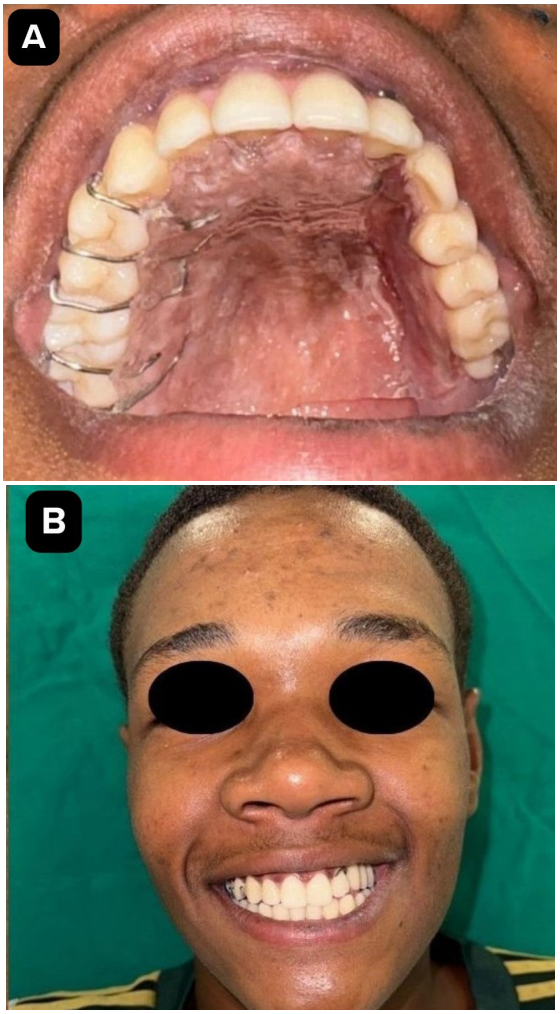

Figure 6 – A: Provisional removable partial denture occlusal view; B: provisional removable partial denture frontal view

Discussion

OM is primarily a slow-growing odontogenic tumour of the mandible, although the present case occurred in the maxilla with rapid progression. However, when OM arises in the maxilla, it can expand into the maxillary sinus, often being diagnosed late after reaching considerable size [10]. It may also involve the palate, orbit and nasal cavity,

causing symptoms related to these structures, as observed in this case. Nevertheless, despite the lesion affecting these areas, the patient did not exhibit any associated symptoms at any point [10].

Regarding imaging features, OM can present as a unilocular lesion or a destructive lesion with poorly defined borders and a multilocular growth pattern, characterised by thin residual bone trabeculae arranged perpendicularly, giving the typical "tennis racket" appearance [1], as described in this case report. CBCT is an indispensable tool for determining the precise location of the tumour and its relationship with adjacent structures, providing a detailed three-dimensional assessment of the lesion and its anatomical interfaces [12]. In the present case, CBCT not only aided in diagnosis, but also ensured the safety of a minimally invasive surgical approach.

Chrcanovic and Gomez [4] conducted a systematic analysis of 1,692 OM cases reported in the literature and described that approximately 75% of lesions exhibited signs of cortical bone perforation, 62.9% presented a multilocular radiographic appearance and 34.7% showed angular septa. Nearly 20% of lesions caused root resorption of adjacent teeth, and 53.8% of cases resulted in tooth displacement and/or impaction due to lesion growth. While the tumour in this case aligned with the predominant features described in the systematic analysis, imaging in this case did not reveal root resorption of adjacent teeth.

Regarding treatment, 44.3% of cases are managed with conservative surgery, including 35.3% treated by enucleation, 8.6% by curettage, and 0.4% by cytoreduction. In 55% of cases, treatment involves resection, with 36.2% undergoing segmental resection and 18.8% undergoing partial or marginal resection [10]. The initial surgical approach may be performed conservatively, with the aim of minimising aesthetic, functional, and psychosocial repercussions [14]. Paediatric patients undergoing conservative treatment should be subjected to continuous and rigorous lifelong monitoring [14]. In the case presented, given the patient's young age, a conservative surgical technique was selected, prioritising the preservation of function and aesthetic harmony.

OM is associated with a high recurrence rate, ranging from 10-43%, with an average of 25%, which can be attributed to its myxomatous nature, lack of encapsulation and penetration into surrounding bone without immediate destruction [11]. According to Chrcanovic and Gomez [4], 42.1% of recurrences occurred within 12 months post-treatment, and 73.7% occurred within 24 months post-treatment. In the present case, the patient remains under follow-up, with no recurrence and a good prognosis.

Conclusion

Although odontogenic myxoma is an uncommon tumour, its aggressive behaviour often leads to mutilating treatments when diagnosed late. Dental surgeons (DS) must therefore recognise its clinical and imaging characteristics for early diagnosis to improve patient prognosis. Additionally, DS must be prepared to work in an inter and multidisciplinary manner to determine the best therapeutic approach. As such, this case highlights the importance of accurate diagnosis of odontogenic myxoma, as well as the need for individualised treatment planning and appropriate rehabilitation, to ultimately promote the patient's quality of life.

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