Case Report

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Odontogenic myxoma of the mandible: a case report

Abdulkarim Khalil, PhD[a], Karam G. Ahmad, MD[a,*], Modar Khalil, MD[c], Rabab Salloum, PhD[b]

Introduction: The odontogenic myxoma is a tumor of the jaws which arises from the mesenchymal portion of the tooth germ, either the dental papilla, the follicle, or the periodontal ligament. It is a slow-growing, painless, nonmetastasizing, central tumor of the jaws, chiefly the mandible. Radiographically, the classic presentation may vary from a unilocular radiolucency to a multilocular lesion with well-defined or diffuse margins. On histological study, it is composed of triangular or stellate connective cells, anastomosed by fine extensions, and embedded in abundant mucoid material.

Case presentation: We present the unusual case of an odontogenic myxoma involving a 37-year-old female patient, which had acquired large dimensions and involved the right half of the mandible, including the ramus; the patient was treated with large resection surgery, with satisfying medium-term results.

Clinical discussion: Early diagnosis of such lesions is very important, as the patient avoids extensive surgical procedures that involve losing a large part of the jawbones and their subsequent impact on the patient's quality of life.

Conclusion: Although there is no fixed treatment plan for the management of odontogenic myxoma, treatment includes surgical management that may range from simple enucleation and curettage to surgical excision; wide surgical resection is appropriate for cases of large size to avoid recurrence.

Keywords: case report, multilocular, myxoma, odontogenic tumor, surgery

Introduction

Odontogenic myxomas account for 3–6% of all odontogenic tumors[1]. According to the World Health Organization's classification of odontogenic tumors in 2005, odontogenic myxomas are rare, benign tumors of ectomesenchymal origin with or without odontogenic epithelium[2]. Previous reports of this tumor have been discussed by many investigators, most recently White and his colleagues, who summarized pertinent data on over 90 individuals and a series of cases of odontogenic myxoma of the jaws, including their group of nine cases. Absolute proof of origin from the odontogenic apparatus is lacking for this lesion, but it appears most likely because of the frequent occurrence of this lesion in the jaws and its almost universal absence in any other bone of the skeleton. For example, in a study by McClure and Dahlin of 6000 cases of bone tumors at the Mayo Clinic up to 1976, no cases involving bones other than the jaws were found[3].

HIGHLIGHTS

- Attention to any enlargement of the maxillofacial region, even if it is not painful for the patient.
- Quick guidance for the patient to conduct the clinical, radiological, and histological examinations required to reach the appropriate diagnosis and perform the indicated surgical procedure as soon as possible.
- A periodic clinical and radiological examination for a relatively long period is highly recommended because of the high recurrence potential present with such lesions.

Their clinical and radiological manifestations are variable and nonspecific and often lead to confusion with other benign and malignant lesions. A range of clinical and radiological arguments, supported by histological examination, is necessary for an accurate diagnosis and an appropriate treatment strategy[4]. Care must be used in distinguishing the odontogenic myxoma histologically from a variety of other myxoid lesions, including myxoid neurofibroma, myxoid liposarcoma, and myxoid chondrosarcoma. The treatment of odontogenic myxomas is surgical excision, followed by cautery. Extensive lesions may require resection to eradicate the tumor. Although this is a benign neoplasm, it frequently exhibits insidious local invasion, making its complete removal difficult, a problem augmented by the loose, gelatinous nature of the tissue itself. The prognosis is good despite unpredictable recurrence. The tumor is not sensitive to X-ray radiation. A frankly malignant form of this tumor, an odontogenic myxosarcoma, is known but is exceedingly rare[3]. The purpose of our work is to present a case of large odontogenic myxoma in the right half of the mandible, including the ramus.

This case report has been reported in line with the SCARE (Surgical CAse REport) criteria 2020[5].

[a]Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, [b]Faculty of Medicine, Department of Histopathology, Tishreen University Hospital, Latakia and [c]Faculty of Medicine, Department of Otorhinolaryngology, Al Mouwasat University Hospital, Damascus, Syria

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*Corresponding author. Address: Germany, 26789. Tel.: +017643961841. E-mail: karam93ahmad@gmail.com (K.G. Ahmad).

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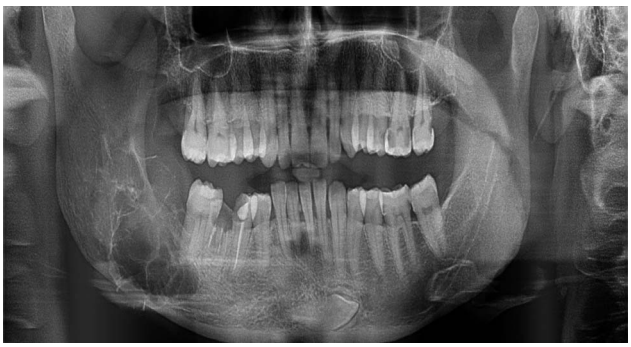

Figure 1. Preoperative panoramic radiograph displays a soap bubble appearance of the right posterior mandible, encompassing the molar region and extending to the ramus.

Case presentation

A 37-year-old female presented to the oral and maxillofacial surgery department with a painless mild expansion of the right posterior mandible and gradually increasing swelling for 6 months. There was no history of trauma or infection. The past medical and dental history was noncontributory. Extraoral examination revealed a swelling measuring ~3 cm in size, extending from the right mouth commissure to the corner of the mandible and inferiorly to the lower border of the mandible. The color, texture, and temperature of the overlying skin were normal and there was no complaint of paresthesia. Intraoral examination showed a firm, nontender swelling with lingual plate expansion involving the molar and premolar region. The overlying mucosa was normal. A panoramic radiograph displayed a large well-defined multilocular radiolucent lesion with a soap bubble appearance on the right posterior mandible. There was an impacted right lower canine out of the surgical field (Fig. 1).

Some tumors that can be suggested based on radiological appearance include odontogenic keratocyst, ameloblastoma, central giant cell granuloma, aneurysmal bone cyst, intraosseous haemangioma, etc[6–8]. About the multilocular appearance in the current case, ameloblastoma was considered a differential diagnosis. A final diagnosis can only be arrived at based on clinical, radiological features and histopathology.

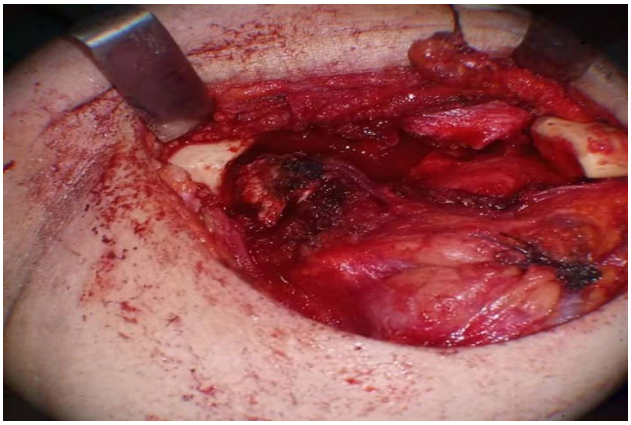

Figure 2. Segmental resection of the posterior mandible with a safety margin of 1 cm.

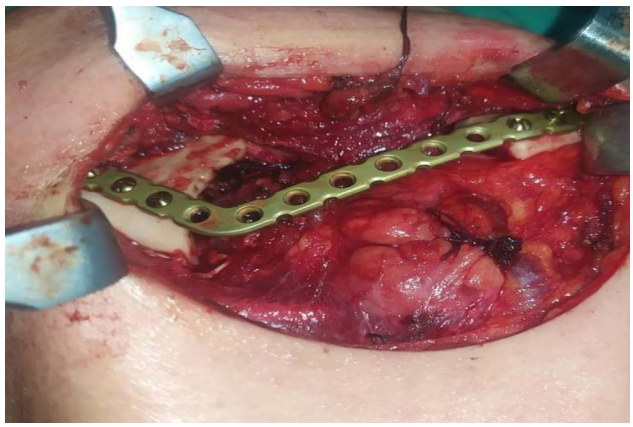

Figure 3. Reconstruction plate with fixation screws after resection of the tumor with safety margins.

A segmental resection of the posterior mandible was planned under general anesthesia. Under sterile and aseptic conditions, a submandibular approach was performed. The entire lateral surface of the mandibular ramus and body was exposed. Segmental resection of the posterior mandible was performed with a safety margin of 1 cm (Fig. 2) and a reconstruction plate was applied (Fig. 3). Maxillomandibular fixation was maintained for 30 days using intermaxillary screws. The patient was discharged from the hospital after 5 days and the postoperative course was without complications.

The specimen was sent for pathological examination (Fig. 4). A loose arrangement of spindle-shaped stellate cells was seen during the pathological examination, which confirmed an odontogenic myxoma (Fig. 5). Post-surgical clinical and radiographic follow-up was done at 1 week, 4 months, 7 months, and 14 months (Fig. 6A–D). There was no evidence of recurrence observed during the follow-up period.

Discussion

Thoma and Goldman first described odontogenic myxoma (OM) in 1947. OM usually occurs in the second and third decade of life

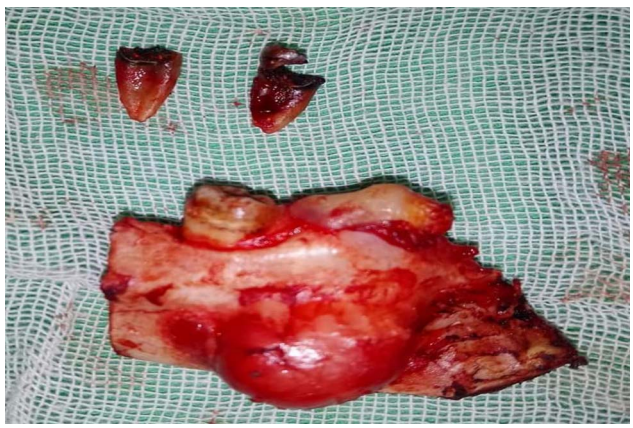

Figure 4. Lingual view for the gross specimen.

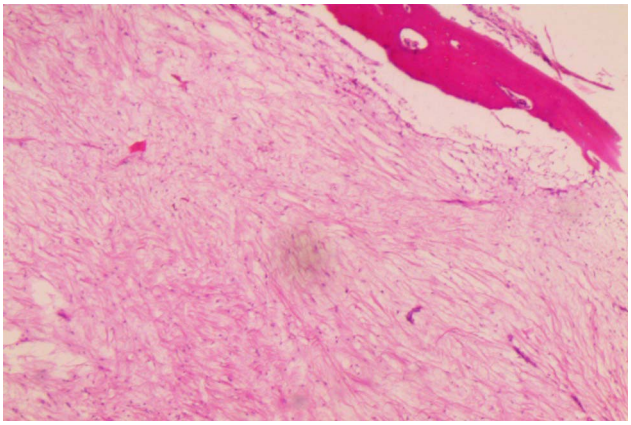

Figure 5. Stellate and spindle-shaped nuclei within a loose, myxoid stroma (hematoxylin and eosin ×40).

in young adults with marked female predilection, this concurs with our patient, who was 37 years old. Most of these tumors occur in the mandible[9]. OM is considered a slow-growing, nonmetastasizing tumor characterized by asymptomatic expansion of the jaw, as was the case for our patient. Its aggressive nature causes bone perforation, root resorption, tooth displacement, and mobility. Production of a mucoid ground substance by the stellate tumor cells causes rapid growth of the tumor[10]. These tumors usually show variable radiographic features ranging from small unilocular lesions to large multilocular lesions. Our case presented with a multilocular radiolucency with the presence of fine, angular septa visible on the panoramic radiograph involving the right aspect of the mandible extending to the inferior half of the ramus approximately.

To this day, there is no consensus for the management of OM. According to several authors, the recommended treatment is surgery, either by a radical resection (segmental or block resection, hemimandibulectomy), or a conservative approach (enucleation, curettage, and marginal resection), depending on the size of the tumor[11]. In our case, the segmental resection was the therapeutic option according to the large size of the tumor and to reduce the possibility of the next recurrence, and with a follow-up of 14 months, we did not notice any recurrence. Reconstruction will be delayed until an adequate disease-free period has passed.

Conclusion

Although OM is a rare lesion, it should not be overlooked. The definitive diagnosis of myxoma is based on histological examination, although clinical and radiographic data may be sufficient to diagnose it. We can suggest a radical excision in case of large tumors where it is difficult to remove this tumor in a conservative manner.

Ethical approval

This case report did not require review by the Ethics Committee of Tishreen University Hospital, Latakia, Syria.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of

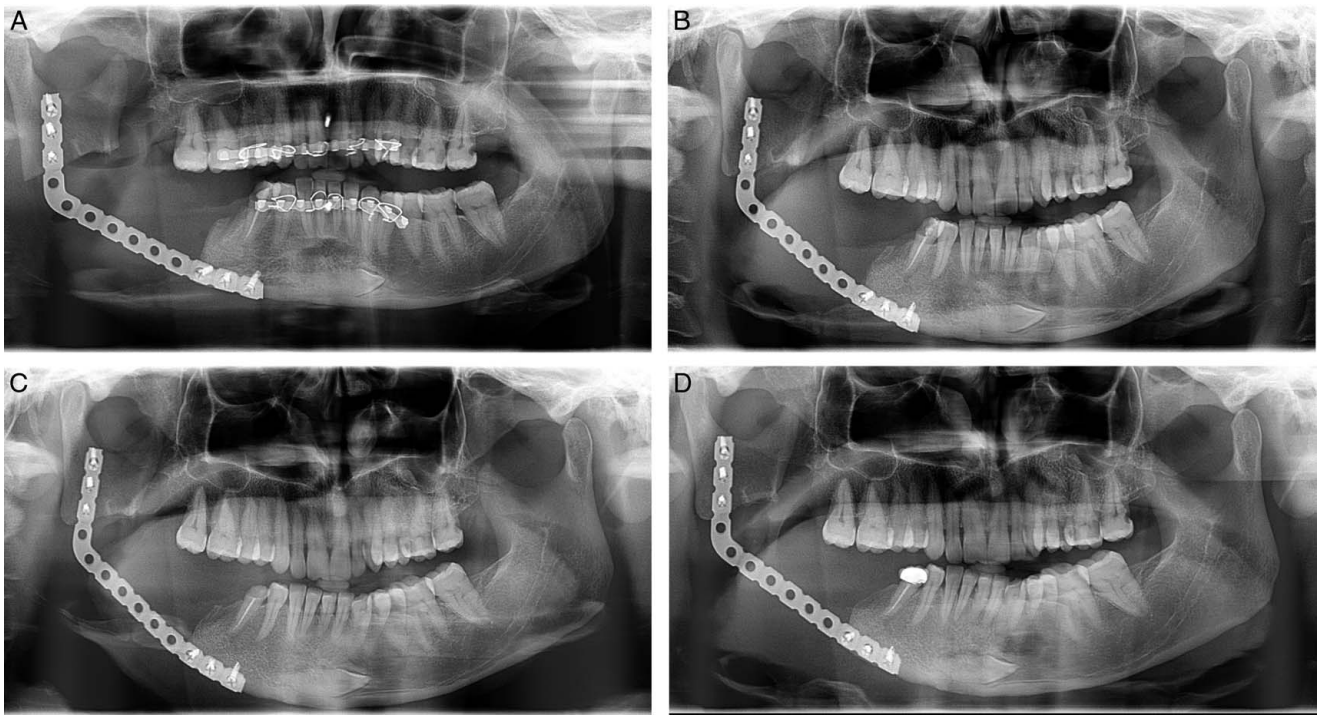

Figure 6. (A) One week postoperative panoramic radiograph of the mandible reconstructed. (B) Four months postoperative panoramic radiograph. (C) Seven months postoperative panoramic radiograph. (D) Fourteen months postoperative panoramic radiograph.

the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

A.K.: data interpretation and as a mentor and reviewer for this case report; K.G.A.: study concept, collection of the data, drafting, literature review, and editing of the manuscript; M.K.: writing the paper; R.S.: histological examination and writing the paper.

Conflicts of interest disclosure

All the authors declare that they have no conflicts of interest.

Research registration unique identifying number (UIN)

This is not an original research project involving human participants in an interventional or observational study but a case report; this registration was not required.

Guarantor

Karam G. Ahmad.

Data availability statement

The data are available for sharing.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Referencing the checklist

This case report has been reported in line with the SCARE Criteria.

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