Follicular Ameloblastoma in the Mandible- A Case Report

Dr. Santha Devy Arumugam[1,*] Dr. Bhuvanesh Kuppusami[2] Dr. Umamaheswari Giri[3] Dr. Vidyalakshmi . S.[4] Dr. Joice Vinodine C[5] Dr.Ijjas Fahi[6]

Professor and Head, Department of Oral & Maxillofacial Pathology and Oral Microbiology, Indira Gandhi Institute of Dental Sciences, Sri Balaji Vidyapeeth Deemed to be University, Puducherry, India.
Senior Lecturer, Department of Oral & Maxillofacial Pathology and Oral Microbiology Sri Venkateshwaraa Dental College, Ariyur,Puducherry, India.
Senior Lecturer, Department of Oral & Maxillofacial Pathology and Oral Microbiology, Indira Gandhi Institute of Dental Sciences, Sri Balaji Vidyapeeth Deemed to be University,Puducherry, India.
Professor, Department of Oral & Maxillofacial Pathology and Oral Microbiology, Indira Gandhi Institute of Dental Sciences, Sri Balaji Vidyapeeth Deemed to be University,Puducherry, India.
Senior Lecturer, Department of Oral & Maxillofacial Pathology and Oral Microbiology Indira Gandhi Institute of Dental Sciences, Sri Balaji Vidyapeeth Deemed to be University, Puducherry, India
MDS., Post Graduate student, Department of Oral & Maxillofacial Pathology and Oral Microbiology, Indira Gandhi Institute of Dental Sciences,Sri Balaji Vidyapeeth Deemed to be University,Puducherry, India.
*** Corresponding Author:** Dr. Santha Devy Arumugam, Professor and Head, Department of Oral & Maxillofacial Pathology and Oral Microbiology,Indira Gandhi Institute of Dental Sciences, Sri Balaji Vidyapeeth Deemed to be University,Puducherry, India.

ABSTRACT

Odontogenic jaw tumors are a group of lesions originating from epithelium remnants associated with teeth development. Most odontogenic lesions occur in the posterior mandible, followed by the posterior maxilla; they rarely occur in the anterior region. These tumors are asymptomatic and they can be associated with an unerupted tooth or may displace developing teeth. Ameloblastomas are slow-growing, locally invasive tumors affecting the maxillofacial region. There are various histologic variants of ameloblastoma. The most common is follicular ameloblastoma. Follicular ameloblastoma is commonly seen in elder people. Follicular ameloblastomas have got high recurrence rate. The radiographic finding shows a multilocular radiolucency with well-defined borders. This paper presents a case of follicular ameloblastoma in a female patient in her early 40s with swelling on the right mandible region.

INTRODUCTION

Ameloblastoma is also known as adamantinoma or adamantoblastoma. Malassez introduced the term adamantinoma. Ameloblastoma is derived from two words, “amel” and “blastos, " meaning enamel and germ, respectively.[1] Robinson gave one of the most accepted definitions of ameloblastoma as a tumor that is usually unicentric, nonfunctional, intermittent in growth, anatomically benign, and clinically persistent.[2] Ameloblastomas are tumors of odontogenic epithelium. Ameloblastoma accounts for 1% of all oral tumors and 11% of all odontogenic tumors.[3] Ameloblastomas are encountered in patients across a wide age range. Ameloblastoma occurs in individuals in the middle to older age groups. Ameloblastoma begins as a central lesion of the bone which is slowly destructive but tends to expand bone rather than perforate it. The overlying mucosa appears normal. It is frequently discovered during routine dental examinations. Based on histological type, ameloblastoma may be classified into follicular, plexiform, acanthomatous, basal cell pattern, unicystic, granular cell pattern, papilliferous ameloblastoma, hemangioma ameloblastoma, desmoplastic, plexiform unicystic, clear cell, dentinoameloblastoma, melanoameloblastoma, and keratoameloblastoma.[4] The most common histologic variants of ameloblastoma are follicular and plexiform variants of ameloblastoma. They both comprise of 27.7% and 21.1%, respectively.[5].

CASE PRESENTATION

A female patient in her early 40s had a chief complaint of swelling and pain in the lower right back tooth region for 4 months [Figure 1]. History of the present illness revealed that the patient was asymptomatic 4 months back when a swelling appeared in the mandibular right posterior region. Initially, the swelling was smaller in size, but it gradually increased to the present size. Difficulty during mastication. No history of trauma and pus discharge from that region was noted. On extraoral examination, a well-defined swelling of 5x5cm is present on the right facial region. Superior-inferiorly, the swelling extended 1.5 cm from the tragus of the ear to 0.5 cm to the inferior border of the mandible. Antero-posteriorly, the swelling extended 1 cm from the corner of the mouth to the angle of the mandible. The overlying surface of the swelling was smooth and of normal skin color. On intra-oral examination, diffuse swelling is evident over the right lower back tooth region extending super-inferiorly from the Maxillary tuberosity to the retromolar region and

medio laterally from pterygomandibular raphe to the mandibular ramus, pus discharge noted in the 46 distal sites.[Figure-2]

INVESTIGATIONS

The orthopantomogram revealed an image with well-defined demarcated borders of approximately 6x6cm and several radio-opaque flecks towards the medial periphery. The lesion extends from the coronoid process without involving the sigmoid notch, till the body of the mandible. Tiny bony structures are seen in the distal border of the lesion [Figure 3]. Before an excisional CT image was taken, a Computed tomography scan showed a coronal image that revealed the buccolingual expansion of the cortical plate, which is evidence of a breach in the lesion. There is no evidence of internal septae formation. [Figure 4]. Based on history, clinical examinations, and radiology examination a provisional diagnosis of ameloblastoma in the right mandibular region was given.

DIFFERENTIAL DIAGNOSIS

Radiological differential diagnosis Odontogenic keratocyst, odontogenic myxoma, central giant cell granuloma, and Brown's tumor of hyperparathyroidism.

TREATMENT

Under the aseptic condition, oro-endotracheal intubation was done and general anesthesia was administered to the patient. segmental resection of the mandible was done under general anesthesia concerning the left lateral incisor up to the left mandibular angle region. The bony defect was reconstructed with a locking reconstruction plate to give reasonable cosmetic and functional outcomes to the patient. Layer-wise closure was done, and pressure dressing was given. The whole resected segment was sent for histopathologic examination [Figure-5]. The histopathological examination of the biopsy specimen revealed the presence of follicles that consist of a core of loosely arranged angular cells resembling the stellate reticulum of an enamel organ. A single layer of tall columnar ameloblast-like cells had been found surrounding the

central core. [Figure-6a] The nuclei of these cells were located at the opposite pole to the basement membrane & reversed polarity with few follicles revealing microcyst formation [Figure-6b]. These findings were strongly suggestive of follicular ameloblastoma.

OUTCOME AND FOLLOW-UP

The patient was stable and well-oriented at the time of discharge. The patient's aesthetics and function were restored. The patient was followed up for 6 months with no evidence of complication or recurrence [Figure-7].

DISCUSSION

Ameloblastomas are rare, representing about 1% of all jaw tumors. They typically manifest in individuals between the third and fifth decades of life. Approximately 80% of ameloblastoma cases occur in the mandible, with the majority (70%) located in the ramus. These tumors can vary significantly in size, ranging from 1 to 16 cm, and can lead to facial asymmetry, tooth displacement, malocclusion, and pathological fractures. In the presented case, the patient exhibited a substantial hard swelling in the mandible's ascending ramus and molar region, resulting in facial asymmetry.

Ameloblastomas arise from epithelial cellular elements and dental tissues at various stages of development. Although they occur with equal frequency in both genders, there is a slight predominance in women, as observed in this case. The majority of mandibular ameloblastomas (70%) are located in the molar ramus region, with 10-15% associated with unerupted teeth. In the current case, the ameloblastoma was situated in the molar ramus region without any association with unerupted teeth.

Ameloblastomas are classified into two main types: extraosseous (peripheral) and intraosseous. Peripheral ameloblastomas typically present as a slow-growing mass that is confined to the gingiva or alveolar mucosa, without involving the underlying bone. Intraosseous ameloblastomas are further categorized into unicystic, desmoplastic, mixed cystic, and solid types. The mixed cystic and solid forms are more aggressive and have a higher likelihood of recurrence. Histopathologically, ameloblastomas can be classified into various types including follicular, plexiform, acanthomatous,

and granular cell types. Less common variants include desmoplastic, basal cell, clear cell ameloblastoma, keratoameloblastoma, and papiliferous ameloblastoma. Among these, the plexiform type is less aggressive and has a lower recurrence rate. [6,7]

Ameloblastomas generally present as a painless, slow-growing hard mass. In this case, the patient experienced a painless, hard swelling that developed over two years before symptoms appeared. Other clinical symptoms may include pain or anesthesia in the affected area. The patient in this case also reported paresthesia over the left cheek region. According to Becelli et al., about 38.3% of patients with mandibular ameloblastoma present with swelling, 13.3% with paresthesia of the mandibular nerve, and 10% with altered occlusion of the teeth. Radiographically, ameloblastomas appear as radiolucent lesions that can be either unilocular or multilocular, with or without expansion of the cortical plate.[8]

Treatment of ameloblastomas can be categorized into three main modalities: conservative approaches (such as enucleation and curettage or marsupialization) and radical surgery (such as resection with or without continuity defect). For solid-multicystic ameloblastomas of the mandible, a resection approximately 1.5–2 cm beyond the radiological limits is recommended to ensure complete removal of all cysts. In this case, a marginal clearance of 2 cm was achieved, with histopathological examination revealing it to be of the follicular variety. The patient underwent a hemimandibulectomy with simultaneous reconstruction to preserve aesthetics. [9]

The likelihood of recurrence is influenced by the surgical treatment method. Annual follow-up for at least 10 years is generally recommended, with some authors suggesting annual follow-up for 5 years, followed by biannual follow-up for up to 25 years. The patient in this case remains under follow-up with no signs of recurrence to date.

LEARNING POINTS/TAKE HOME MESSAGES

- **Priority in Differential Diagnosis**: When evaluating any swelling in the mandibular posterior region of middle-aged individuals, ameloblastoma should be the foremost consideration in the differential diagnosis.

- **Imaging Requirements**: For small lesions, conventional radiographs typically suffice.

However, for larger lesions, advanced imaging techniques such as CT scans are necessary to facilitate better surgical planning and management.

•**Recurrence Risk and Surgical Approach**: Multilocular or solid ameloblastomas have the highest recurrence rates among all types. Therefore, it is crucial to perform a wide resection of the lesion, including a safety margin of healthy bone, to minimize the risk of recurrence.

•**Surgical Reconstruction**: Resection combined with simultaneous reconstruction using various grafts is essential not only for restoring the patient's aesthetic appearance but also for maintaining the functional integrity of the jaw.

REFERENCES

1.Kumaresan R, Kotrashetti SM. Management of large mandibular ameloblastoma: A case report. J Dentofac Sci 2013;2:21-5.

2. Gupta A, Jindal C. Hybrid ameloblastoma: Report of a rare case and review of literature. Int J Oral Maxillofac Pathol 2011;2:68-72.

3. Hasan K, Alok A, Singhal P, Rohatgi N. Plexiform ameloblastoma: A case report and a brief review. MAVEN 2018;2:9-14.

4. Karjodkar FR, editor. Essentials of Oral and Maxillofacial Radiology. 2nd ed. New Delhi. Jaypee Brothers Medical Publishers (P) Ltd.; 2019. p. 403-53.

5. Eversole LR, Leider AS, Hansen LS. Ameloblastomas with pronounced desmoplasia. J Oral Maxillofac Surg 1984;42:735-40.

6. Kahairi A, Ahmad RL, Islah Wan L, et al. Management of large mandibular ameloblastoma—a case report and literature reviews. Arch Orofac Sci 2008;2013:52–5.

7. Varkhede A, Tupkari JV, Mandale MS, et al. Plexiform ameloblastoma of mandible—case report. J Clin Exp Dent 2010;2013:e146–8.

8. Gumgum S, Hogoren B. Clinical and radiologic behavior of ameloblastoma in 4 cases. *J Can Dent Assoc* 2005;2013:481–4.

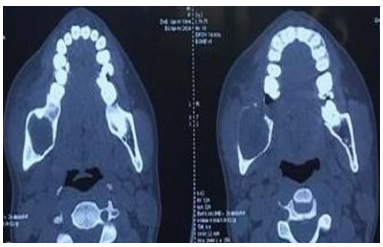
9. Cankurtaran Ceylan Z, Chiosea Simon I, Leonjr BE, et al. Ameloblastoma and dentigerous cyst associated with impacted mandibular 3rd molar teeth. Radiographics 2010;2013:1415–20

FIGURE CAPTIONS

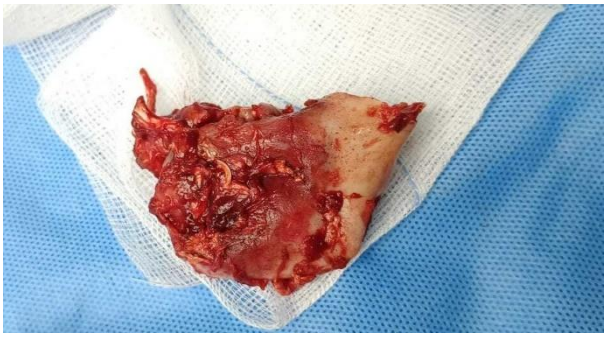
Caption: Figure 1: Extraoral photograph of the patient, revealing a diffuse swelling over the right side of the face

Caption: Figure 2 Intraoral photograph revealing a diffuse swelling from tooth 44 to the retromolar region.

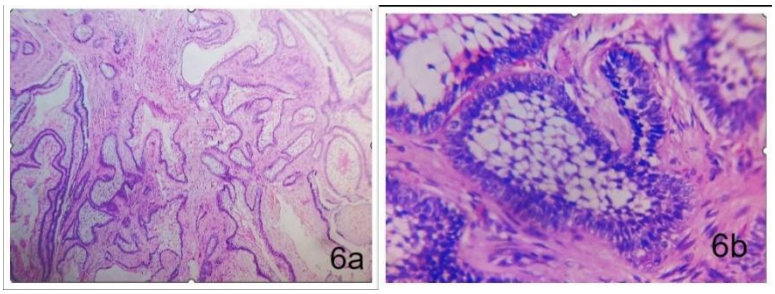
Caption: Figure 3: Radiographic image reveals well-defined demarcated borders of size approximately 6x6cm, and several radio-opaque flecks towards the medial periphery.



Caption: Figure 4: Computed tomography shows a coronal image that reveals the buccolingual expansion of the cortical plate with evidence of a breach in the lesion.



Caption: Figure 5: Surgically excised specimen (hemimandible).



Caption: Figure 6a: The 4x view shows islands of odontogenic epithelium seen in variable size in the form of cords & follicles proliferated in the connective tissue.

Caption: Figure 6b: The 40x view shows peripheral cells which are tall columnar with reversal polarity of the nucleus. The central area exhibits star-shaped stellate reticulum-like cells.

Caption: Figure 7: The patient was followed up for 6 months with no evidence of complication or recurrence.

PATIENT'S PERSPECTIVE

Initially, I felt it was found a swelling in the upper left back tooth region. I visited the

dental hospital and they asked me for some history of swelling in the mouth. After that, they told me to take X-ray photos and fluid samples to identify the disease. The dentist said that it require immediate attention for the problem. They suggested doing surgical excision and explained about surgery. I accepted surgery, and after the surgery, I feel good and without any discomfort. they give some antibiotics and analgesics for 7 days. They do follow-up and regular checkups for till dates in the hospital.