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Case Report

Follicular ameloblastoma: a case series highlighting diagnostic and therapeutic challenges

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ABSTRACT:
Ameloblastomas are rare, odontogenic tumors, thought to be composed of the epithelium of ectodermal origin. Ameloblastomas represent about 1% of all jaw tumors, but they are the second-most common odontogenic tumor.The vast majority of ameloblastomas are benign and slow-growing, with locally aggressive behavior, which can lead to significant pathology and require extensive surgical treatment. According to the WHO Classification of Head and Neck Tumors (2017), there are 7 distinct histological subtypes, each displaying varying biological behaviours. Follicular ameloblastoma is the most common subtype of conventional ameloblastoma that is characterized by a unique histopathological appearance. Follicular ameloblastoma is commonly seen in elderly individuals and have got a high recurrence rate. Most of them occur in the mandible, predominantly in the posterior mandibular region. Microscopically, the epithelial cells are arranged in islands or follicles surrounded by connective tissue. This paper presents four clinical cases of conventional follicular ameloblastoma, highlighting their clinical, radiological, and histopathological characteristics to enhance understanding and aid in timely diagnosis and management of this condition.
Keywords: Ameloblastoma, follicular ameloblastoma, mandible and soap bubble appearance

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INTRODUCTON

Ameloblastomas are rare, odontogenic tumors, thought to be composed of the epithelium of ectodermal origin, which means they are tumors arising from the cells around the tooth root, or in close approximation, derived from the ectoderm germ layer. They represent about 1% of all jaw tumors, but they are the second-most common odontogenic tumor.[1]Ameloblastoma most commonly occurs in the mandible than the maxilla, the ratio being 5:1.The average age of occurrence reported is about 38.9 years.[2]Ameloblastomas can occur at any location in the mandible or maxilla, but the regions of the mandibular molars and ramus are the most prevalent anatomical locations (80%).[3]They are typically slow-growing, painless lesions that have a high recurrence rate and the potential to spread to other parts of the body. Due to their aggressive nature, ameloblastomas can become large lesions causing facial disfigurement, dental malocclusion, and loosening of the teeth. In the recent classification of the World Health Organization (WHO) 2017, ameloblastoma has been classified clinically as conventional, unicystic and peripheral variant, whereas histopathologically under conventional, it has been further classified as follicular and plexiform (being most common) and others (granular, basal cell variant, acanthomatous, clear cell variant etc.)[4] According to the study conducted on 104 ameloblastomas by Shigeru Ueno DDS et al, the mandibular ameloblastomas were of the follicular type in 60 cases (62%) and the plexiform type in 37 cases (38%).

CASE REPORT 1

A 26 year old male patient presented at the department of Oral medicine and Radiology with a diffuse painless swelling on left lower jaw for past 6 months [figure 1]. History of the presenting illness revealed that the patient was asymptomatic 6 months back then a swelling appeared in the mandibular left posterior region. No history of change in the size of the swelling or associated symptoms like pain/ bleeding/ pus discharge. He did not report any incidents of trauma. History of extraction of 37 due to mobility about 6 months back. On extraoral examination, a diffuse nontender swelling with firm to hard in consistency noted on the left mandibular posterior region .The overlying skin appeared normal without any surface alterations or colour change. Non tender and fixed left level IB lymphnodes were noted.

Intraoral examination showed a large lobular non tender swelling of variable consistency (predominantly bony hard with occasional areas of softness) over left mandibular posterior region extending from mesial aspect of 36. Posterior and superoinferior extension was not discernible. Surface mucosa appears mildly hyperkeratotic in relation to edentulous region 37 and 38. An erythematous area over the mucosa was seen due to repeated trauma from 26. Bucco-lingual cortical plate expansion was present, exhibiting no secondary changes [Figure 2]. Grade II mobility with tender on percussion on 36 and edentulous region in relation to 37 and 38 noted.

The orthopantamogram revealed a well defined multilocular radiolucency, involving left mandibular body, angle, ramus, coronoid and condylar process [Figure 3]. CECT scan [figure 4] displayed a large lobular expansile lesion (soap bubble appearance ~ 83 x 56 mm) seen involving left hemi-mandible (condyle, coronoid process, left ramus and body) with cortical erosion involving lingual as well as buccal cortices. The lesion shows peripheral sclerotic rim and is seen to indent left submandibular gland medially, left masseter laterally and is seen to extend postero-superiorly indenting left parotid gland as well as left lateral pterygoid and left infra-zygomatic temporalis muscles. Enlarged left level IB, IIA/B, III, VA nodes seen.

An incisional biopsy was done under local anaesthesia at the site of left posterior alveolar ridge. 3 specimens were taken and sent for histopathological analysis, which revealed: a follicular ameloblastoma predominantly composed of numerous ameloblastomatous follicles with moderately collagenous connective tissue stroma [Figure 5].

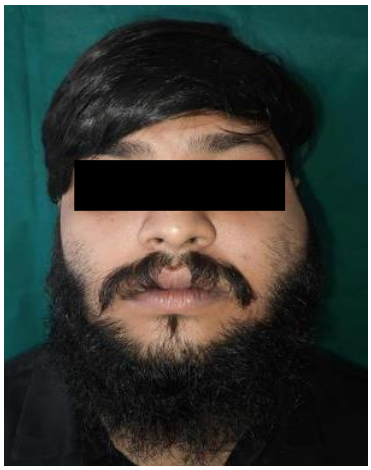


Figure 1. Extraoral photograph of the patient, revealing a diffuse swelling over the left side of face

Figure 2. Intraoral photograph showing a diffuse swelling over edentulous 37 and 38 region

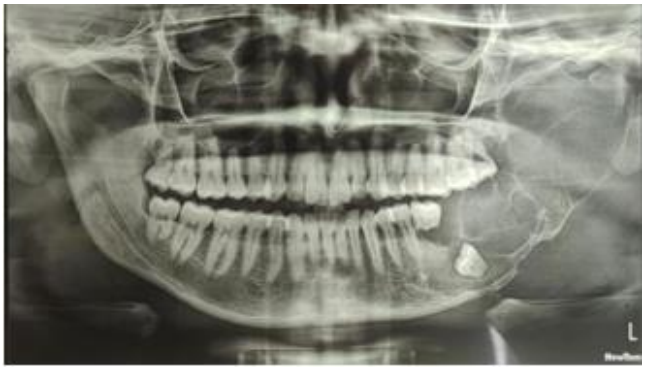


Figure 3. OPG revealed a well defined multilocular radiolucency involving left mandibular body, angle, ramus, coronoid and condylar process.

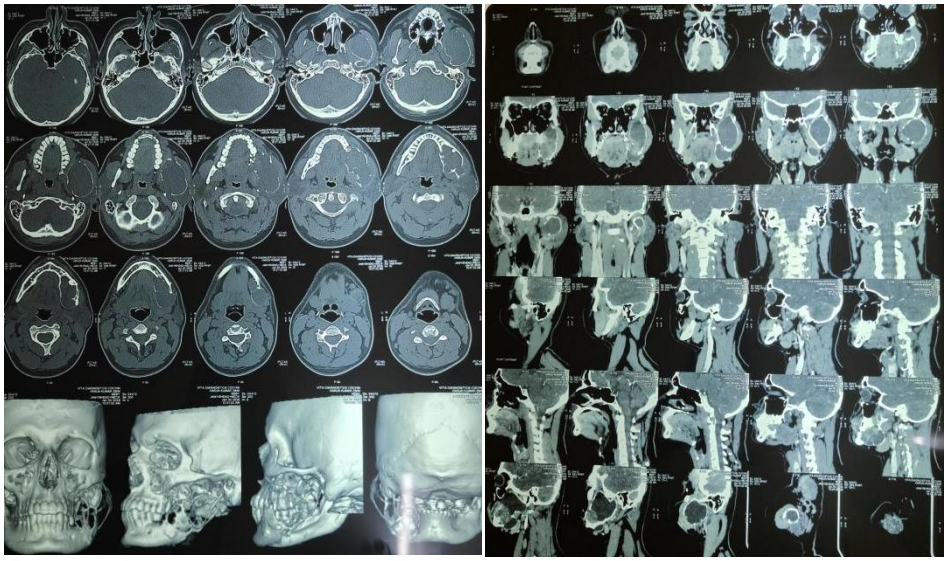


Figure 4. CECT scan showing a large lobular expansile lesion involving left hemi-mandible.

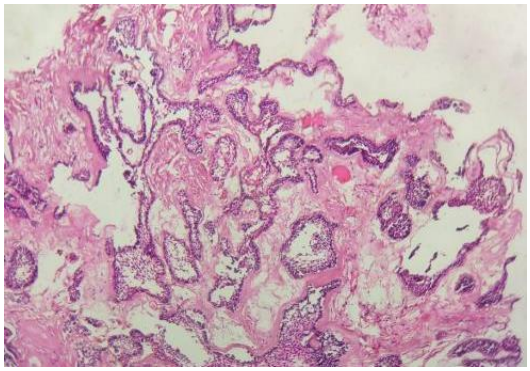


Figure 5. Histological slide confirmed the diagnosis of a follicular ameloblastoma. (H and E :10x)

CASE REPORT 2

A 50 year old female with chief complaint of swelling on the left side of face since past 4 month reported at Oral Medicine and Radiology OPD. History of the presenting illness revealed that the patient was asymptomatic 4 months back then a swelling appeared in the mandibular left posterior region. Initially swelling was small in size and gradually increased to current state. History of pus discharge from lesion since past 2 weeks. No history of other associated symptoms like pain/ bleeding/parasthesia. She did report an incidents of trauma (log hit on face) 6 months back. She is under medications for diabetes mellitus. History of extraction of left lower back tooth due to mobility about 6 months back. Her family and personal histories yielded no patient information. On extraoral examination, a diffuse nontender swelling with firm to hard in consistency noted on the left mandibular body region .The overlying skin appeared normal without any surface alterations or colour change. No cervical lymphadenopathy was noted.

Intraorally, a well defined non fluctuant, non compressible, tender hard swelling was observed, extending from the distal aspect of 33 to 37, affecting both buccal and lingual cortical plates. Obliteration of buccal vestibule noted with pus discharge. Clinically missing 34 and 35 is noted along with supra-eruption of 36 [Figure 6].

Radiographic examination showed a well defined, multilocular expansile hypodense lesion of approx. Size 2.6 x 4.2 x 2.6 cm in the left mandibular symphysis, parasymphysis and body region. The lesion extends anteroposteriorly for periapical aspect of 31 to the distal aspect of 37, superoinferiorly from the crestal region to 6 mm above inferior border of mandible in relation to edentulous 36 region. Breach in the buccal cortical plate were also noted at some points. Bucco-lingual expansion of cortical plate were noted with minimal lingual expansion. Root resorption of apical 1/3rd of 33 were noted. Loss of lamina dura noted in relation to mesial root of 37. Inferior alveolar nerve canal and mental foramen could not trace [Figure 7 and 8]. A fine needle aspiration was ordered which yielded brown colored fluid. An incisional biopsy was performed under local anaesthesia and histopathological analysis showed follicular ameloblastoma [Figure 9].

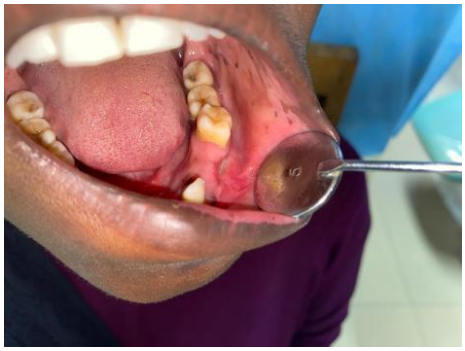


Figure 6. Intraoral photograph showing vestibular obliteration with pus discharge over 34, 35 and 36. region.

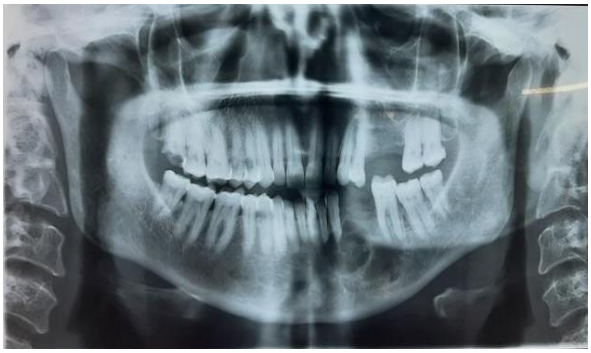
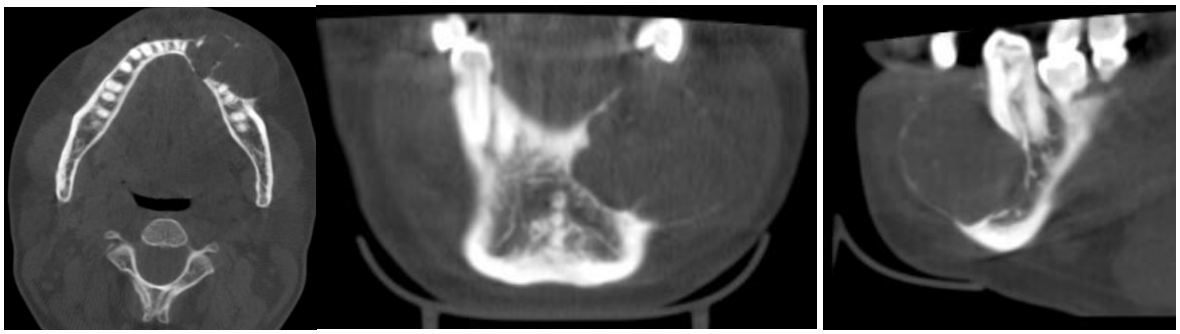


Figure 7. OPG revealed a well defined multilocular radiolucency involving left mandibular body.



(a) (b) (c)

Figure 8. CBCT scan showing a large multilocular expansile lesion involving left hemi-mandible .(a) - axial view; (b) - coronal view; (c) - sagittal view

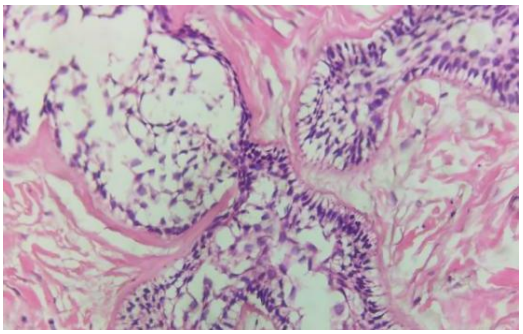


Figure 9. Histopathological view showingnumerous ameloblastomatous follicles with densely collagenous connective tissue stroma (H and E : 40x)

CASE REPORT 3

A 56 year old male patient presented with a swelling on right side of face that had been developing over a period of 1 year [Figure 10]. History of the presenting illness revealed that the patient was asymptomatic 1 year back then a swelling appeared in the mandibular right posterior region. Initially, swelling was smaller in size, but gradually increased to the present size. He did report incident of trauma 1 year back . History of bleeding, parasthesia and pus discharge from that region was noted. History of mobility of right lower back teeth were also given. The patient was under medications for hypertension. Personal history yielded cigarette smoking for past 25 years and betel leaf, areca nut, slaked lime and tobacco chewing for past 4 years.

On extraoral examination, a well defined, mildly tender swelling of approximate size 5 x 6 cm with variable consistency noted on the right mandibular posterior region. Intraoral examination showed a large well defined mildly tender swelling of variable consistency over right mandibular posterior region [Figure 11]. Surface mucosa appears erythematous. Buccal cortical plate expansion was present from 43 to 48. Lingual cortical plate expansion also noted in relation to 47 and 48, exhibiting no secondary changes. Grade 3 mobile 48; grade 2 mobile 45 and 46; grade 1 mobile 44 was noted. Clinically missing 47 were noted.

Orthopantamography (OPG) revealed a well defined, multilocular radiolucency of characteristic honey comb appearance measuring approximately 6 x 4 cm in the right mandibular angle, body and

parasymphysis region [Figure 12]. CBCT scan revealed a large, multilocular expansile hypodense lesion of approx. Size 5.9 x 4.8 x 4.5 cm in the right mandibular angle, body and parasymphysis region [Figure 13]. The lesion extends anteroposteriorly from periapical aspect of 43 to angle region. Expansion and breach in the buccal and lingual cortical plate was noted. Root resorption of apical 1/3rd of 44, 45, 46 and 48 was noted. Inferior alveolar nerve canal and mental foramen could not trace.

A fine needle aspiration was ordered which yielded reddish-brown colored fluid. Histopathological examination of the biopsy specimen revealed a follicular ameloblastoma predominantly composed of numerous ameloblastomatous follicles with densely collagenous connective tissue stroma [Figure 14].

Figure 10. Extraoral photograph of the patient, showing a diffuse swelling over the right side of face

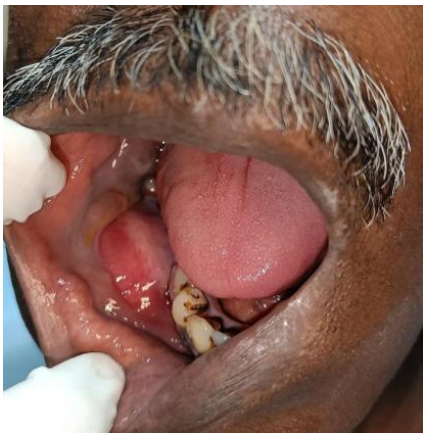


Figure 11. Intraoral photograph showing a swelling in relation to right mandibular body region.

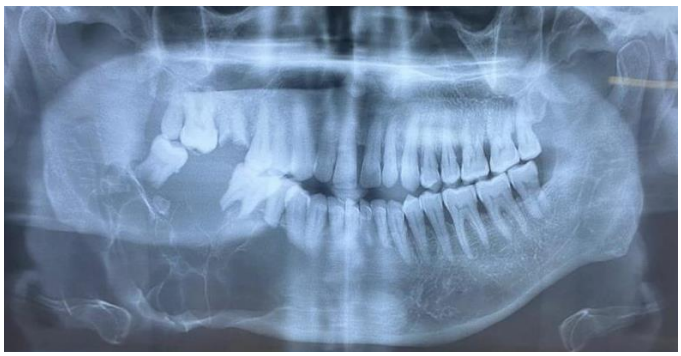
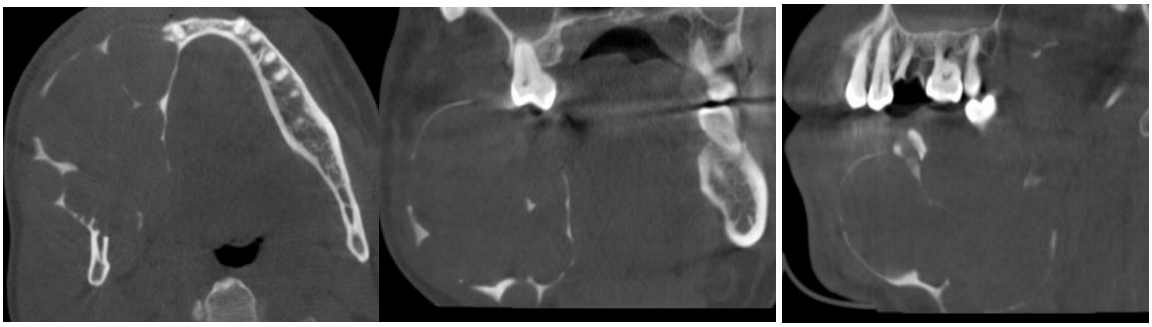


Figure 12. OPG revealed a well defined multilocular radiolucency extending from distal aspect of 48 to the periapical aspect of 43.



(a) (b) (c)

Figure 13. CBCT scan showing a large multilocular expansile lesion involving right hemi-mandible .(a) - axial view; (b) - coronal view; (c) - sagittal view

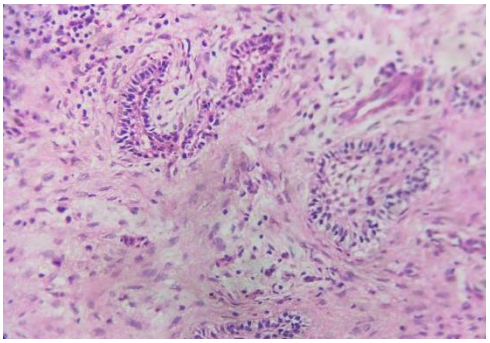


Figure 14. Histopathological view showinga follicular ameloblastoma predominantly composed of numerous ameloblastomatous follicles with densely collagenous connective tissue stroma (H and E : 40x)

CASE REPORT 4

A 39 year old male Patient complains of pain on the left side of the lower jaw for 4 monthsreported at Oral Medicine and Radiology OPD [Figure 15]. History of presenting illness revealed that the patient was apparently normal 4 months back when he noticed a dull aching pain on lower left jaw. Patient is under medication for Seizure with last episode on December 2023. He was diagnosed for megaloblastic anemia in 2004 for which he was under medication.Patient had extraction of lower left posterior teeth 15 years back.

A diffuse swelling and facial asymmetry noted extraorally on left side. On intraoral examination, indentation of upper teeth noted on residual alveolar ridge in relation to 37, 38, with mild surface hyperkeratosis. A non tender, diffuse swelling noted on left residual ridge mucosaof approximate size 1.5 x 1.3 cm irt 37, 38 extending from distal aspect of 36 till the retromolar region. Mild vestibular obliteration noted irt 37, 38 region. Surface appears to be smooth and soft cystic in consistency in the lingual aspect of 37, 38 region, superiorly bony hard in consistency and firm in consistency towards the retromolar region [Figure 16].

Radiographic examination shows a well-defined multilocular hypodense area of approximate size 3 x 2.4x 3.5cm extending anteroposteriorly from the distal aspect of 43 to the distal aspect of 36. Expansion and extensive destruction of buccal cortical plate present and thinning and focal area of destruction of lingual cortical plate noted at the level of 36. Root resorption noted irt 42,31,32,33,34,35,36. Divergence of root noted in relation to 33, 34 [Figure 17]. An incisional biopsy was performed under local anaesthesia at the site of left body of mandible and histopathological analysis revealed a follicular ameloblastoma [figure 18].

Figure 15. Extraoral photograph of the patient, showing a diffuse swelling over the left side of face

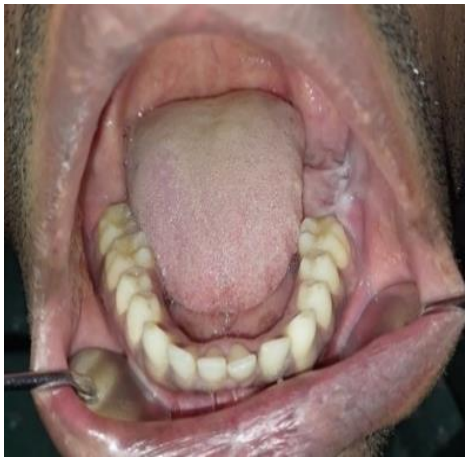
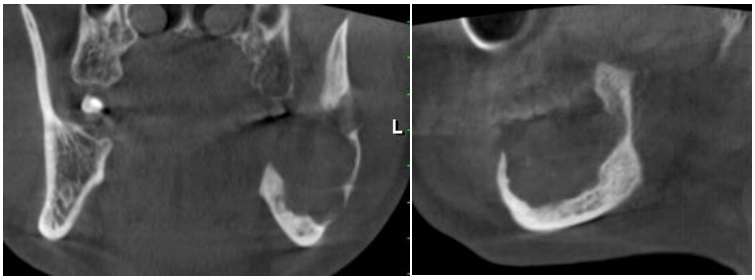


Figure 16. Intraoral photograph showing a diffuse swelling over 36, 37 and 38 region.



(a) **(b)**

Figure 17. CBCT scan showing a large multilocular expansile lesion involving left hemi-mandible . (a) - coronal view; (b) - sagittal view

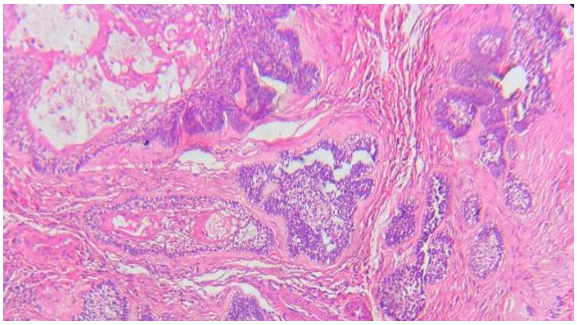


Figure 18. Histopathological view showing a follicular ameloblastoma predominantly composed of numerous ameloblastomatous follicles with densely collagenous connective tissue stroma (H and E : 40x)

DISCUSSION

Ameloblastoma which is also known as adamantinoma or adamantoblastoma is a benign but locally aggressive odontogenic tumor that arises from remnants of the enamel organ, dental lamina, or basal cells of the oral epithelium.It is the most common epithelial odontogenic tumor, comprising 1% of tumors and cysts arising in the jaws.[5]In 1868 Broca first described this tumour, Churchill in 1934 coined the term ameloblastoma.[6]It was derived from the English word "amel," which means enamel, and the Greek word "blastos," which means the germ. Robinson in 1937 described ameloblastoma as a benign tumor that is usually "unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent."[7] Most common etiologic factors of ameloblastoma include trauma, inflammation, chronic irritation, and infection with history of traumatic injury or extraction.[8]

Roughly 80% of ameloblastomas occur in the mandible, with a distribution of 70% in the ascending ramus–molar region, 20% in the premolar region, and 10% in the anterior mandible.[9]Our series showed a predominance of mandibular involvement, consistent with existing literature.An association with unerupted teeth is observed in approximately 10–15% of cases. Although isolated reports describe a male predominance with a ratio of 2:1, most studies fail to confirm a statistically significant gender predilection.[10] The typical age of onset ranges from 20 to 50 years, with an average age of 27.18 years.[11] The cases in our series varied in age (26 to 56 years), gender, location, duration, and symptoms, highlighting diverse clinical presentation of this condition.

Clinically, ameloblastoma is most often characterized by an indolent, painless swelling that progressively enlarges, frequently giving rise to facial asymmetry. Expansion of the cortical plates—buccal and/or lingual—is a common finding, with potential extension into adjacent soft tissue structures. Less frequently, clinical manifestations may encompass malocclusion, displacement and mobility of teeth, mucosal ulceration, periodontal pathology, and sensory disturbances such as parasthesia within the affected region.[12] Radiographically, ameloblastoma most often manifests as a well-circumscribed unilocular or multilocular radiolucency, occasionally displaying a characteristic honeycomb or soap-bubble pattern. The lesion is frequently associated with cortical plate thinning or expansion, scalloped margins, and in some instances, cortical perforation. In more advanced stages, resorption of adjacent dental roots is a common finding.

Due to its slow-growing nature, the diagnosis of ameloblastoma is often delayed. Additionally, its clinical presentation can resemble other mandibular tumors, which further complicates accurate diagnosis.[13,14] A definitive diagnosis of ameloblastoma can only be established through histopathological examination. The follicular variant is typified by round, oval, or irregular epithelial islands, closely resembling the enamel organ. The islands display peripheral palisading of tall columnar cells with reverse nuclear polarity, wherein the nuclei are oriented away from the basement membrane. Centrally, the epithelial component consists of angular

cells that simulate the stellate reticulum of the developing tooth germ. These epithelial aggregates are delineated by intervening mature fibrous connective tissue stroma.

The treatment of ameloblastoma primarily involves surgical excision due to its aggressive nature and high recurrence risk. Wide local excision or segmental resection with 1–2 cm margins is the treatment of choice, significantly reducing recurrence. Marginal resection may be considered for smaller, well-demarcated lesions, though with a slightly higher risk of recurrence.

Adjunctive measures such as cryotherapy or application of Carnoy's solution may be used to lower recurrence following conservative procedures, though their role is limited. Radiotherapy and chemotherapy are not routinely indicated. Long-term follow-up (5–10 years) with periodic imaging is essential to monitor for recurrence.

CONCLUSION

Ameloblastomas, though benign, exhibit locally aggressive behavior with a significant potential for recurrence if not adequately managed. The present case series highlights the classical clinical, radiographic, and histopathological features of follicular ameloblastoma, reinforcing the need for careful diagnostic evaluation and timely surgical intervention. Wide surgical excision with adequate margins remains the most effective treatment approach, while long-term follow-up is indispensable to detect recurrence at an early stage. Through these cases, the importance of early recognition, comprehensive imaging, and histopathological confirmation is emphasized to ensure optimal patient outcomes and to minimize the functional and esthetic morbidity associated with this tumor.

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