SVOA Dentistry

ISSN: 2753-9172

ScienceVolks Open Access

Case Report

Plexiform Variant of Ameloblastoma in Body of Mandible: Case Report and Review of Literature

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DOI: https://doi.org/10.58624/SVOADE.2024.05.0183

***Received:** July 19, 2024* ***Published:** August 12, 2024*

Abstract

Ameloblastoma is a benign but locally aggressive odontogenic epithelial neoplasm arising from remnants of dental lamina and dental organ [1,4]. This neoplasm is more prevalent between 2nd to 4th decade of life and mandible (11%) is more commonly involved than maxilla (1%) [2,5]. It has an aggressive behavior and recurrent course, It is rarely metastatic. Ameloblastoma represents 10% of odontogenic tumors and 1% of all tumors and cysts involving the maxillo mandibular region [1]. The World Health Organization (WHO) classification of 2017 describes ameloblastomas of the following four types: Solid/multicystic ameloblastoma (91%) unicystic ameloblastoma (6%); extraosseous/peripheral ameloblastoma (2%); and metastasizing ameloblastoma (1%). [3,8] A conclusive diagnosis can only be made by histopathology because it has wide range of histologic patterns and has radiographic characteristics with other lesions such the odontogenic keratocyst, giant cell tumour, aneurysmal bone cyst, and renal cell carcinoma metastasis.[5] Treatment for each type of ameloblastoma differs depending on its biological behaviour and rate of recurrence. This report presents a case of ameloblastoma involving the body of mandible with resorption of the root apices of canine, premolars and first molar. The lesion extends till the base of mandible with intact lower border. This was surgically excised preserving the lower border of mandible with safe sound margins of bone.

***Keywords:** Ameloblastoma; Odontogenic tumors; Mandible*

Introduction

Ameloblastoma is described by Robinson (1937) as a benign tumor that is 'usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent. [1] It was first recognized by Cusack in 1827 [2] and designated as an 'adamantinoma' in 1885 by the French physician Louis-Charles Malassez in 1930[4,8]. It was rechristened 'ameloblastoma' by Ivey and Churchill [3,8].

They may occur at any age, even though nearly half of the tumors do occur between the ages of 20 and 40 years with equal sex predilection [5]. It is known to induce root resorption and manifests radiographically as an expansile radiolucency with thinning and perforated cortices. This is the most common neoplasm affecting the jaws, yet only accounts for 1% of all tumors of the maxilla and mandible and 11% of all odontogenic tumors. It is most common in the mandible (nearly 80% of cases) where in 10% are located in the anterior mandibular region, 20% are in the premolar region, and 70% percent are in the molar or ascending ramus. In maxilla predominantly found in the maxillary tuberosity, canine or molar region. (nearly 20% of cases) [5].

The tumour is usually asymptomatic and slowly growing in nature so remains unnoticed until causes large destruction presenting a large facial swelling.

Ameloblastomas are notorious for their propensity to recur and for their invasive behavior. Small incipient lesions may be misinterpreted for a typical periapical granuloma or cyst, in which case the tooth may be removed or treated endodontically while the lesion remains unidentified or receives insufficient care. Since this lesion needs to be treated more severely than other benign periapical lesions. This case report presents a study of a plexiform ameloblastoma with excisional biopsy and continuous follow up.

Case Presentation

A 17-year-old male patient reported in outpatient department of Oral and maxillofacial surgery at Peoples Dental Academy (PDA) with chief complaints of swelling in his left lower third of face and loosening of teeth for 4 months. The patient initially noticed a swelling on his left side of the mandible 2 years back, which showed slow growth and progressive nature with no history of trauma. The swelling was painless. On extraoral examination there was notable facial asymmetry and a diffuse swelling in the left mandibular region extending from 2 cm away from the corner of mouth till 2 cm anterior to the angle of mandible and a the level of corner of mouth to the base of the mandible (Fig 1). Skin over swelling appeared normal in color and texture. On palpation swelling was firm in consistency, non-tender and immobile without sign of paresthesia on the affected side. The temperature of skin over swelling was normal. On Intraoral examination, vestibular obliteration was from 33 to 36 region. Buccal cortical plate expansion was noticed. The area was slightly tender and the tooth 33,34,35,36 had Grade-1 mobility. On palpation swelling was soft in consistency and eggshell crackling was noted. This is suggestive of a swelling which has caused resorption of buccal cortical plate.

Fig 1: *A firm swelling in the area of left mandibular body region.*

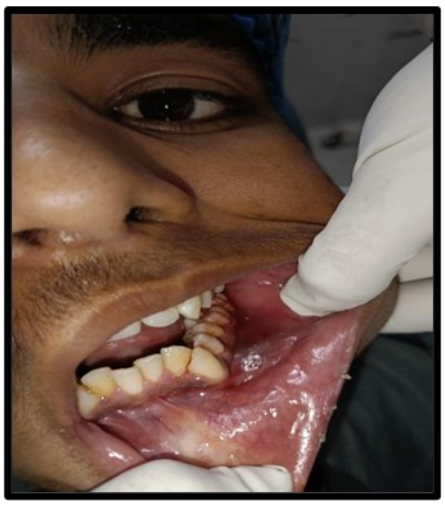

Fig 2: *Vestibular obliteration present.*

On aspiration, from the lesion showed blood-tinged fluid with inconclusive diagnosis.

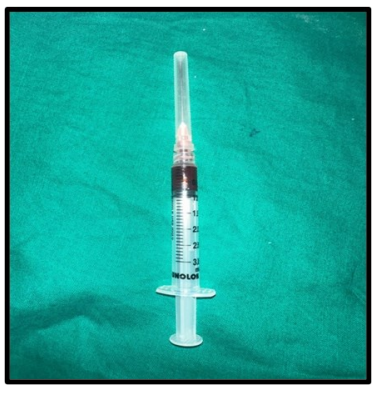

Fig 3: *Aspiration*

Radiographically, solitary well defined radiolucent lesion involving body of mandible with resorption root of mandibular canine, premolars and first molar. The lesion extends till the base of the mandible with intact inferior border. (Fig. 3). The medical history was not significant, and the patient was in good general health

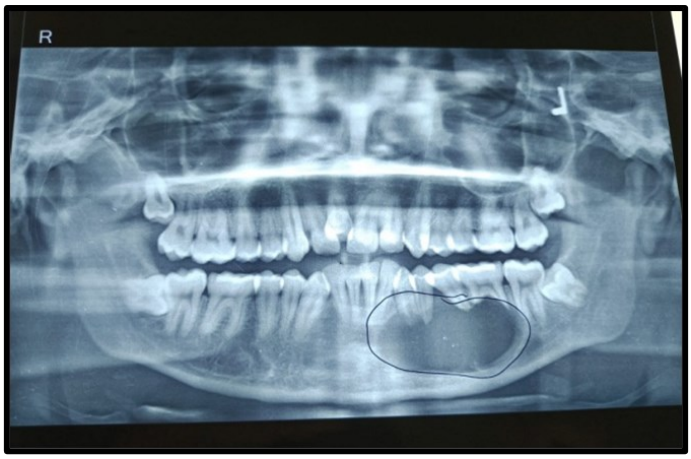

Fig 4: *Radiograph showing well defined unilocular radiolucency with resorption of roots of canine, premolar and molar.*

Based on clinical and radiographic evaluation diagnosis of cystic lesion was made.

Differential diagnosis: - Unicystic Ameloblastoma (Root resorption of teeth)

Excisional biopsy was done under local anesthesia and sent for histopathological examination using intraoral approach followed by extraction of 33,34,35,36.

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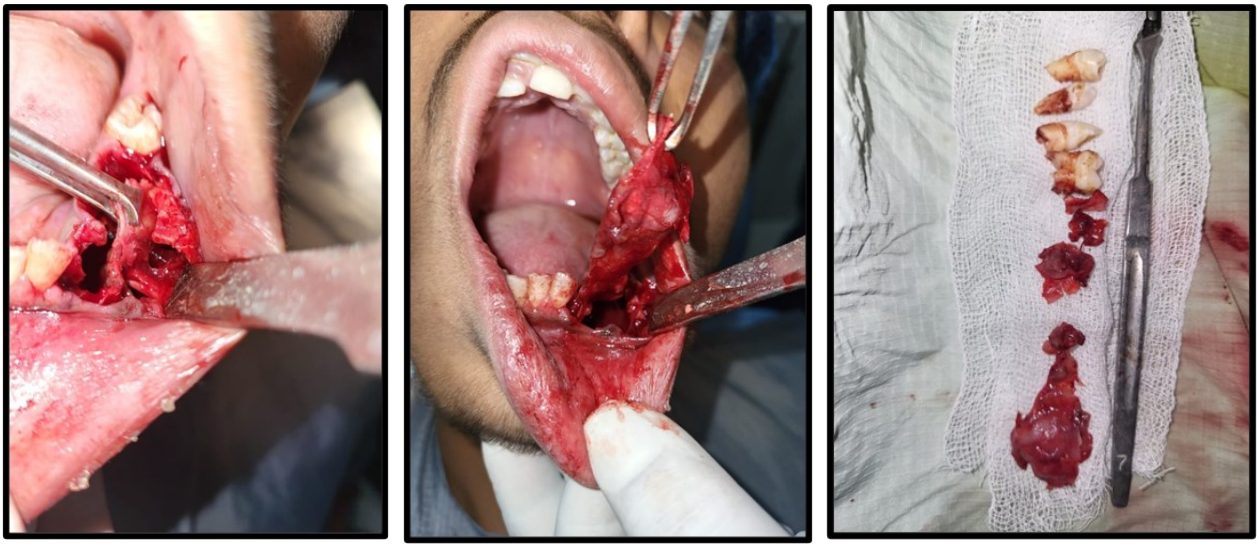

Fig 5: *Intraoperative pictures showing complete enucleation of the lesion with extraction of involving tooth under local anesthesia.*

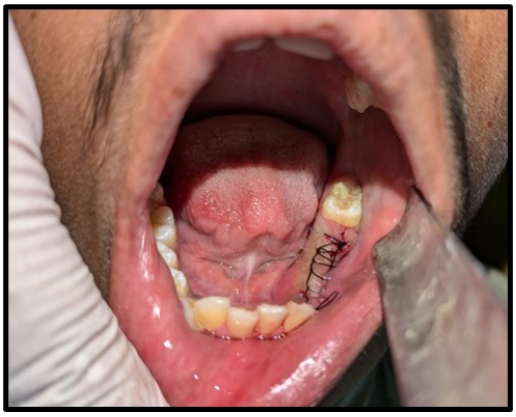

Fig 6: *Closure after enucleation.*

The pathology report confirmed the diagnosis of a plexiform ameloblastoma. The microscopic findings of the specimen revealed nests and anastomosing trabeculae of odontogenic epithelium in fibro connective tissue with foci of squamous metaplasia. No evidence of malignancy seen.

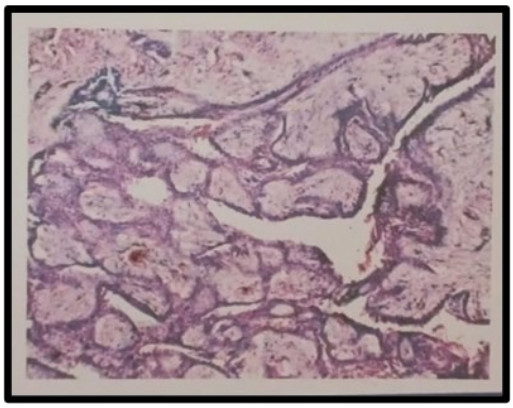

Fig 7: *Histological slide confirmed the diagnosis of a plexiform ameloblastoma.*

No complications occurred intraoperatively. Antimicrobial and anti-inflammatory drugs were prescribed for the 1st post-operative week. At the first control appointment, the patient presented a good healing and reduced inflammation. The patient was scheduled for monthly follow up.

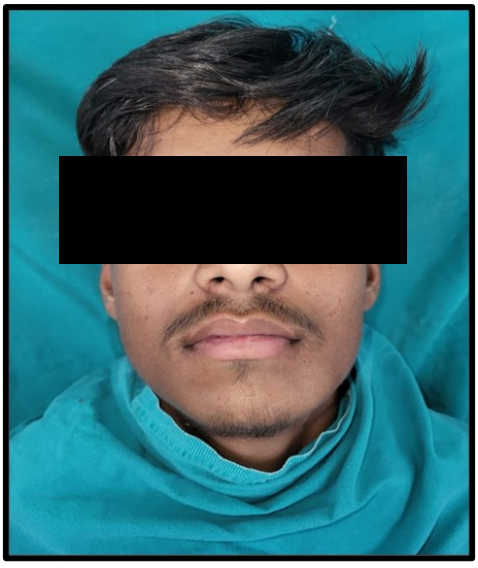

Fig 8: *Postoperative picture after 1 month marked reduction in swelling can be noticed.*

Discussion

Ameloblastoma is a benign epithelial odontogenic tumor which is slow growing but is aggressive and destructive in nature causing erosion of bone and invading adjacent structures. [8]. Initially the plexiform variant was thought to be hyperplastic epithelial growth of the cystic lining rather than a true ameloblastoma since it didn't meet the published histologic criteria by Vickers and Gorlin in 2007. It was Gardner who discovered the plexiform variant of ameloblastoma in 1981. [5] Mutations in genes that belong to the MAPK pathway are found in many ameloblastomas, the most common being the BRAFV600E mutation [8].

In mandible 10% percent ameloblastomas are located in the anterior region of the mandible, 20% percent are in the premolar region, and 70% percent are in the molar or ascending ramus area.10 to 15% of ameloblastomas are linked to a tooth that has not yet erupted. [2,3,5]

Clinically it manifests as a painless swelling with facial deformity, malocclusion, ulceration and paresthesia of affected area [2,4].

Radiologically, ameloblastomas are known to exhibit unilocular or multilocular radiolucency, giving the appearance of soap bubbles or honeycombs. In advance stages expansion of cortical plates with scalloped margins and perforations with resorption of the involved teeth can be seen. Radiographically, differential diagnosis can be odontogenic keratocyst, aneurysmal bone cyst, fibrosarcoma, or a giant cell tumor. [2] Since none of the radiological features are pathognomonic, the definite diagnosis of ameloblastoma is only established by biopsy. [8]

Histologically, ameloblastoma is classified into follicular, acanthomatous, granular cell, basal cell, and plexiform. Follicular and plexiform ameloblastomas are the most common, with incidence rates of 27.7% and 21.1% respectively, followed by acanthomatous and the granular types [5].

The appearance of odontogenic epithelium anastomosing islands in contrast to a follicular pattern is referred to as "plexiform." The cords or sheets of epithelium are bounded by columnar or cuboidal ameloblast like cells surrounding more loosely arranged epithelial cells. The supporting stroma is loosely arranged and vascular. There is a notable decrease in the aggressiveness and recurrence rate of the plexiform pattern [3,4] Also, plexiform ameloblastomas are found to remain in the primitive stage of tumour differentiation, whereas follicular and a canthamatous types are thought to undergo squamous differentiation [3].

Generally, maxillary ameloblastomas are more aggressive and prone to higher recurrence than mandibular, mainly due to the thin maxillary cortical bone that provides a weak barrier for the local spread of the tumor. [3,8]

Treatment may vary from being conservative to radial resection depending upon the extent of the lesion and its presentation. Various therapeutic options consist of marginal resection, aggressive resection, marsupialization followed by enucleation, and enucleation followed by chemical cauterization with Carnoy's solution [5]. Extensive resection can result in functional deficit, cosmetic defects which may causes psychological depression in patients.

Conservative surgery, such as marsupialization, enucleation, curettage, enucleation mixed with Carnoy's solution, enucleation combined with curettage, and curettage combined with cryotherapy, has also been tried in an attempt to prevent these complications. In essence, many surgeons believe that the benign and slow-growing nature of the tumor may permit initial “conservative” management, with “radical "salvage surgery performed on an as-needed basis if the tumor recurs. [9] According to Huffman and Thatcher, the diagnosis of ameloblastoma should not indicate immediate jaw resection. [12] Instead, they recommend treatment being based on the clinical features of the tumor, including slow growth, tendency to not invade soft tissues, and the lack of wide dissemination. “Complete removal of the tumor with preservation of the inferior border of the mandible whenever possible. [9] Although reoccurrence rate up to 40 % has been documented by the conservative methods.

In our case we conservatively enucleated the lesion because radiographically it presented as an unilocular lesion with well corticated margins, so we opted to be conservative and keep the patient with regular follow up every 6 monthly.

Conclusion

Ameloblastoma is an aggressive odontogenic neoplasm of an enamel organ. The decision of treatment is determined by the extent of the lesion and invasion into surrounding structures. Histology plays a major part which predicts the aggressiveness of the lesion, increasing the recurrence. The prognosis is favorable if identified earlier. Recurrences often present after 15 years or more. Therefore, it is important to emphasize the need for long-term periodic follow-ups.

Conflict of Interest

The authors have no conflict of interest to declare.

Funding

No Funding received

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Citation: Jain N, Pillai A, Choubey A, Sharma P, Dubey S, Sharma D. Plexiform Variant of Ameloblastoma in Body of Mandible: Case Report and Review of Literature. *SVOA Dentistry* 2024, 5:4, 141-146. doi:10.58624/SVOADE.2024.05.0183