

Intraluminal Unicystic Ameloblastoma: A Case Report

Atul U. Bhagwat

Registrar, Vaishampayan Memorial Government medical College & Hospital, Solapur 413001, Maharashtra, India.

Correspondence to:

Dr. Atul U. Bhagwat, Registrar, Vaishampayan Memorial Government medical College & Hospital, Solapur 413001, Maharashtra, India.

ABSTRACT

Ameloblastoma is a neoplasm of odontogenic epithelial origin. It is the most common odontogenic neoplasm, except odontoma. Its incidence, combined with its clinical behavior, makes ameloblastoma the most significant odontogenic neoplasm. Unicystic ameloblastoma (UA) refers to those cystic lesions showing clinical, radiographic, or gross features of a mandibular cyst, but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. It accounts for 5-15% of all intraosseous ameloblastomas. We report a case of unicystic ameloblastoma in a 65-year-old male and review the literature.

KEYWORDS: Unicystic Ameloblastoma, Odontogenic Neoplasm, Intraosseous Ameloblastoma

INTRODUCTION

Many benign lesions of odontogenic and non-odontogenic origin can cause mandibular swellings. Odontogenic lesions include ameloblastoma, radicular cyst, dentigerous cyst, keratocystic odontogenic tumour. While central giant cell granuloma, fibro-osseous lesions and osteomas are non-odontogenic lesions.¹ The most common tumour of odontogenic origin is ameloblastoma, which develops from odontogenic epithelial elements and dental tissues in their various phases of development. This tumor has characteristic features like slow-growth, persistent, and locally aggressive neoplasm of epithelial origin with its peak incidence is in the 3rd to 4th decades of life. It is often associated with an unerupted third molar and majority of ameloblastomas arise in the mandible.²

There are three forms of ameloblastomas 1) multicystic 2) peripheral and 3) unicystic tumors.³ Multicystic ameloblastoma is the most common variety contributing 86% of cases. Peripheral tumors are odontogenic tumors that occur solely in the soft tissues covering the tooth-bearing parts of the jaws with the histological characteristics of intraosseous ameloblastoma. Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts.⁴ The goal of treatment ameloblastoma is to achieve complete excision and restoring patient's health.

MATERIALS AND METHODS

We present a case of a unicystic mandibular ameloblastoma in a 65-year-old male Patient complains of pain in the lower front region of the jaw since 2-4 days. Patient apparently asymptomatic 4 months back when he had trauma to lower anterior region. Initially this

was small swelling of peanut size, which was painless, increased gradually to the present size.

Intraoral examination showed a swelling in mandibular anterior region near right corner of mouth, 3X2cm in size, from midline to right corner of mouth & superoinferiorly from vermilion border to 2cm above lower margin of mandible, diffuse borders, normal overlying surface and temperature and soft in consistency. Examination of lesion reveals a soft swelling of present buccally extending from lower 41 to 44 region. Swelling is soft, fluctuant, bluish colour, mobility of teeth associated with lower 42. [Fig-2]



Fig. 1 extra oral examination. Fig.2: intra oral examination: a swelling in mandibular anterior region near right corner of mouth, 3X2 cm in size, soft, fluctuant, bluish colour

A provisional diagnosis of the keratocystic odontogenic tumour was given, and the differential diagnosis was ameloblastoma was given. Aspirate showed brownish fluid. Microscopic examination showed the presence of RBC's.

A panoramic radiograph revealed a well-defined unilocular radiolucency in the anterior region of the mandible from the distal aspect of 33 to 43. [Fig-3] Intraoral periapical radiograph shows well-defined

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radiolucency between roots of lateral incisor and canine. Root displacement of 42, 43 [Fig-4]. Occlusal radiograph shows radiolucent lesion extending from 43 to 33 & also show displacement of roots [Fig-5].

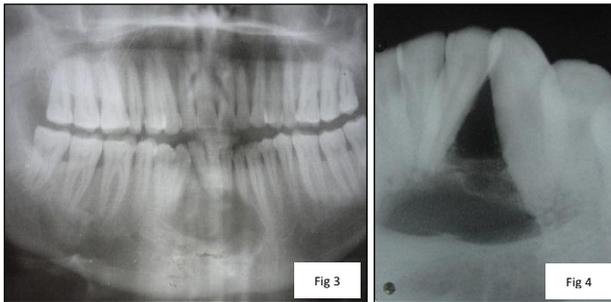


Fig.3: Occlusal radiograph showed radiolucent lesion extending from 43 to 33 along with displacement of roots of 42, 43. Fig.4: Intraoral periapical radiograph shows well defined radiolucency between roots of lateral incisor and canine. Root displacement of 42, 43

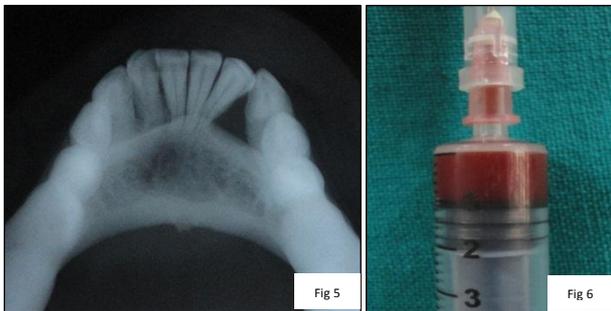


Fig.5: Occlusal radiograph showed radiolucent lesion extending from 43 to 33 along with displacement of roots of 42, 43. Fig.6: Aspiration of cystic lesion showed brownish fluid



Fig.7: Intra operative picture showing complete excision of the lesional tissue. Fig.8: Gross examination of soft tissue of size 4x1.5 cm cystic lining with small growth towards the luminal side.

Aspirate of the cystic lesion was brown in colour [Fig.6]. Excisional biopsy was performed, and gross examination of soft tissue reveals small growth within the lining of cyst towards the luminal surface [Fig.7,8]. Histopathological examination showed fibrous wall with thin bony trabeculae and muscle tissue at the periphery. The fibrous wall was lined by proliferating ameloblastomatous epithelium towards the lumen.

The luminal surface of the wall showed proliferating islands, sheets & interlacing strands of odontogenic epithelium forming the plexiform pattern. Peripheral cells of epithelial islands were cuboidal to columnar & arranged in palisaded fashion. At places, cells were

forming showing the reverse polarity of nuclei and peripheral vacuoles. Inside the islands were loosely arranged stellate reticulum like cells with frequent areas of squamous metaplasia. Areas of stromal degeneration were seen. The peripheral connective tissue wall was fibrous with numerous blood vessels & focal areas of the chronic inflammatory cell infiltrate. One of the two foci of ameloblastomatous epithelium is presenting connective tissue suggestive of mural growth. Overall features were suggestive of “Unicystic Ameloblastoma-Intraluminal variant” [Fig-9, 10].

The enucleation was carried out for this case and patient was under antibiotic coverage with Ofloxacin and Metronidazole and then discontinued after infection control. On follow-up at the intervals of 6 months to 1 year [Fig-11, 12].

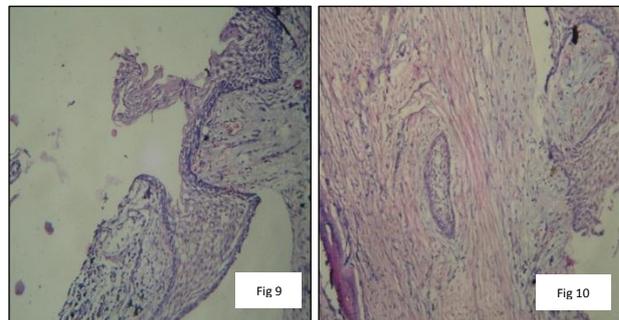


Fig.9 and 10: histopathological examination showed H and E stained (40X) section with luminal surface showed strands of odontogenic epithelium forming plexiform pattern. Peripheral cells of epithelial islands were cuboidal to columnar & arranged in palisaded fashion.

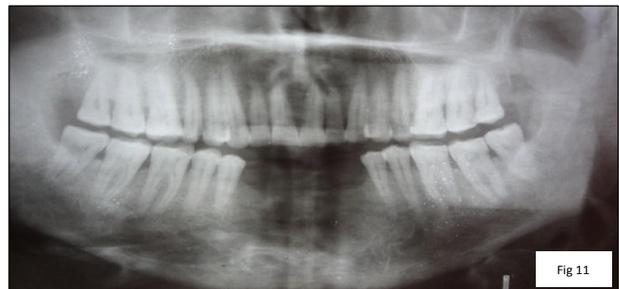


Fig.11 and 12 shows post-operative panoramic radiograph during 6 month and 1yr follow up period respectively.

DISCUSSION

Ameloblastoma is a benign, locally aggressive and infiltrative odontogenic neoplasm which rarely metastasize.¹ They comprise only 1.3% of all jaw cysts and tumours and 2nd most common odontogenic

neoplasm constituting 10 % of neoplasm of odontogenic origin.¹⁻⁴

Cusack was the first to give accurate the description of an ameloblastoma. Later Falkson described ameloblastoma in 1879. Weld was credited for the first histo-pathological description in 1952. Weld called the tumor 'cystsarcoma' but suggested that it could have arised from a tooth bud or the dental lamina.⁵

In 1998, a critical review of 193 cases of UA verified that most of UA can be associated with impacted tooth i.e. Dentigerous variant (n=90) and not associated with impacted tooth i.e. non-dentigerous variant (n=101).⁶

Leider et al., among the cases found 3 pathological mechanisms for UA – (1) reduced enamel epithelium associated with developing tooth, (2) cell rests of odontogenic epithelium in the cyst wall, and (3) a solid ameloblastoma undergoing cystic changes.⁷

Vindhya et al., reported a case of plexiform ameloblastoma and discussed the differential diagnosis of unicystic ameloblastoma with odontogenic cysts [8]. In 1998, three histological subtypes of UA were recognized, based on the character and extent of tumor cell proliferation within the cyst wall⁷:

- Luminal - cyst wall lined by ameloblastic epithelium (Group I).
- Intraluminal - cystic lesion showing proliferation of ameloblastic epithelial lining into the cystic lumen (Group II).
- Mural- cystic lesion with ameloblastic epithelial invasion into the supporting connective tissue or islands of ameloblastic epithelium in either follicular or plexiform pattern Invasive ameloblastoma arising from lining of the cyst.

The predominance of unilocular configuration is exceptionally marked for dentigerous variant i.e. 4.3:1 compared to non-dentigerous variant 1.1:1. The multilocular lesions demonstrate characteristic 'honeycomb' or 'soap bubble' appearance. Resorption of roots of adjacent teeth is seen in 40% - 70% of cases.⁹

Sandeep G et al., reported a case report of unicystic ameloblastoma which presented in a multilocular pattern which needed aggressive management.¹⁰ In our case report the case was unilocular variant (Non- Dentigerous Type).

UA has a good prognosis, and the probable reason is that the UA is generally cystic, well-localized and surrounded

by a fibrous capsule. However, once the tumor has breached the periphery of the capsule, it can easily infiltrate the surrounding cancellous bone resulting in more aggressive behaviour of the lesion.⁷ The recurrence rate following enucleation and curettage is 10-33% and is more likely in the mural variant. Recurrence of UA may be long delayed, and a long-term postoperative follow-up is essential in the proper management of these patients.⁷

CONCLUSION

Unicystic ameloblastoma tends to affect young adolescent/younger patients, but we reported case that was occurred in 7th decade. UA can only be diagnosed based on histopathological features and cannot be predicted on clinical and radiological grounds alone. The examination of the entire lesion through sectioning at varying levels is mandatory for securing the final diagnosis. UA is believed to be less aggressive and should be treated conservatively.

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