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Case Report

Unicystic Ameloblastoma: A Case Report

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ABSTRACT

Ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm, and only odontoma outnumbers it in reported frequency of occurrence. Its incidence, combined with its clinical behavior, makes ameloblastoma the most significant odontogenic neoplasm.

Unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but on histologic examination shows 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 intraosseousameloblastomas.

Unicystic ameloblastoma is a less encountered variant of the ameloblastoma and believed to be less aggressive. Moreover, recurrence of unicysticameloblastoma may be long delayed and a long-term post-operative follow up is essential for proper management of these patients. In this case report we present a uncommon multilocularunicystic ameloblastoma encountered in a 40 year old male patient.

Key Words: Unicystic ameloblastoma, unilocular, multilocular

INTRODUCTION

Ameloblastomas are benign tumors whose importance lies in its potential to grow into enormous size with resulting bone deformity. The most common tumor of odontogenic origin is ameloblastoma, It is a slow-growing, persistent, and locally aggressive neoplasm of epithelial origin.

The Unicystic ameloblastoma is a less encountered variant of the ameloblastoma, referring to those cystic lesions that show clinical and radiographic characteristics of an odontogenic cyst but in histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation. [(1)] This paper illustrates a case of unicystic ameloblastoma of the mandible in a 40-year old male.

CASE REPORT

A 40-year-old male patient presented to the Department of Oral and maxillofacial surgery with swelling on the left side of the face for last four months.

Extra orally a diffuse swelling present in lower left molar region showing asymmetry of face. Intraorallythere was no associated pain, difficulty in opening the

mouth, chewing or articulating. Oral mucosa was normal systemic examination normal. The lesion extended from lower left premolarsto lower left molars with obliteration of buccal sulcus. (Fig 1)The swelling was tender on palpation. Left submandibular lymph nodes were palpable and tender.

An orthopantomogram (Fig2) showed large cystic lesion in the left side of mandible, the swelling was approximately 5 x 8cm in size, extending from superior border of alveolar ridge to1.5 mm above the inferior border of mandible. Osteolytic lesion was multilocular also showed displacement of premolar roots anteriorly.

CBCT report showed that the cysticlesion was confined to the mandible, with loss of both lingual and buccal cortex. (Fig3)

Fig 1 Preoperative Intraoral view showing obliteration of buccal sulcus.

Fig 2 Preoperative OPG showing lesion extension.

Fig 3 CBCT showing lesion extension.

Fig 4 Resected specimen.

Considering the site, age and it being multilocular a differential diagnosis of ameloblastoma / odontogenic keratocyst was considered. Incisional biopsy was done to confirm diagnosis of Unicystic ameloblastoma. Enblock resection without continuity defect was done under general anesthesia and the specimen was subjected to histopathological examination to recheck the diagnosis again. The gross specimen (Fig 4) revealed buccal cortical expansion with thinning of cortical plates. Expansion of the lingual cortical plate was noted. Histologic examination shows a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation. Based on the histopathological findings, diagnosis of unicystic ameloblastoma showing intraluminal and mural proliferation was confirmed.

DISCUSSION

Unicysticameloblastoma, a variant of ameloblastoma was first described by Robinson and Martinez. [(2)] It is a slow-growing, persistent, and locally aggressive neoplasm of epithelial origin. Its peak incidence is in the 3rd to 4th decades of life and has an equal sex distribution. [(3)] More than 90% are found in the mandible usually in the posterior region. The lesion is often asymptomatic, although a large lesion may cause painless swelling of the jaw. [(4)]

There are three forms of ameloblastomas, namely multicystic, peripheral, and unicystic tumors. [(5)] Multicystic ameloblastoma is the most common variety and represents 86% of cases. Peripheral tumors are odontogenic tumors, with the histological characteristics of intraosseous ameloblastoma that occur solely in the soft tissues covering the tooth-bearing parts of the jaws. Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts. [(6)] Gardner in 1984 has pointed out that there is a difference in biological behavior between these lesions. [(7)] It has been widely

stated that these lesions are less aggressive than their solid or multicystic counterparts and should be treated by enucleation or curettage. However, Gardner has pointed out that unicystic ameloblastoma having the capacity to invade the cancellous bone by penetrating and breaching the fibrous wall.

It has been suggested that recurrence following conservative surgery is more likely to occur in the unicystic ameloblastoma therefore these lesions should be treated in the same manner as solid ameloblastomas. [(8)]

Late recurrence following treatment is commonly seen, the average interval for recurrence being 7 years. Recurrence is also related tohistologic subtypes of UA, with those invading the fibrous wall having a rate of 35.7%, but others only 6.7%. [(9)] Recurrence rates are also related to the type of initial treatment. Lau et al [(10)] reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialization followed by enucleation.

Whatever surgical approach the surgeon decides to take, long-term follow-up is mandatory, as recurrence of unicysticameloblastoma may be long delayed. [(10)]

CONCLUSION

Very rarely, we come across a case with presentation of multilocular, non dentigerous variant and unicystic type in the same. This Unicystic variant of ameloblastoma is of aggressive histologic behavior. We can successfully treat such ameloblastoma with conservation of lower border of mandible and maintaining the esthetics according to patient's psychological demand. This approach can be considered as an alternative to total resection.Surgeon should keep in his mind irrespective of surgical approach long-term follow-up is mandatory, as recurrence of Unicystic ameloblastoma may be long delayed.

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