CASE REPORT

Non-dentigerous variant of Unicystic Ameloblastoma – a case report

Jyotismita Thakuria[1], Rajendragouda Patil[2], Simi Thankappan[3], Udita Singh[4]

Abstract

The ameloblastoma is a rare odontogenic neoplasm of the mandible and maxilla. It is primarily seen in adults in the third to fifth decade of life, with equal sex predilection. Ameloblastoma prevalently occurs in the mandibular molar and the ramus areas. Recurrence frequently appears after inadequate treatment. It shares common radiographic features with other lesions such as the giant cell tumor, aneurysmal bone cyst, and renal cell carcinoma metastasis.

This report presents a case of unicystic ameloblastoma which is not associated with an impacted tooth (non-dentigerous variant) involving entire ramus of the mandible.

Keyword: Ameloblastoma; Plexiform; Mandible; Odontogenic.

Introduction

Ameloblastoma is a true neoplasm of odontogenic epithelium which is a persistent and locally invasive tumor; aggressive but benign growth characteristics. It is described by Robinson (1937) as a benign tumor that is 'usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent'[1]. 70% of ameloblastomas develop in the molar-ramus region of the mandible and are occasionally associated with an unerupted third molar teeth.[2] The neoplasm was first described by Cusack in 1827 [3]. Etymologically, the name derives from the old French word ''amel,'' which means enamel, and the Greek word ''blastos,'' meaning germ or bud. Over time, this tumor has been referred to by many different names including ''cystosarcoma,'' ''adamantine epithelioma,'' ''adamantinoma,'and finally ''ameloblastoma'' [4,5].

1. *Post Graduate Student*
2. *Professor and H.O.D.*
3. *Reader*
4. *Senior lecturer*

Department of Oral Medicine and Radiology

Correspondence Address

1. Dr. Jyotismita Thakuria
Department of Oral Medicine and Radiology, Kothiwal Dental College & Research Centre, Moradabad,
Email: jyotismita.thakuria@gmail.com

Radiographically an ameloblastoma can be a unilocular or multilocular radiolucency with a honeycomb or soap bubble appearance. The chief histopathological variants of ameloblastoma are the follicular and plexiform types, followed by the acanthomatous and granular cell types. Uncommon variants include desmoplastic, basal cell, clear cell ameloblastoma, keratoameloblastoma and papilliferous ameloblastoma. The plexiform pattern is less aggressive and has a significantly lower recurrence rate.

Case Report

A 17 year old boy reported to the Department of Oral Medicine and Radiology with complaint of swelling on left side of face since last 1 year. The swelling was initially smaller in size, but gradually increased to attain the present size. There was no history of pain associated with the swelling.

The medical history was unremarkable and the patient was in good general health. Personal history did not have significant findings.

Extraoral clinical examination revealed notable facial asymmetry with a large, diffuse, smooth- surfaced, swelling on left side of face measuring approximately 4x3 cm with normal overlying skin. It extends from the zygomatic region to the inferior border of mandible superoinferiorly, and from the corner of mouth to the angle of mandible anteroposteriorly. (fig.1)

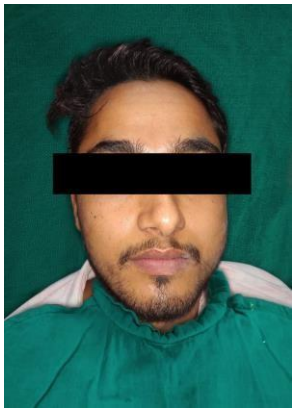

Fig.1

On palpation, the inspection findings of size, and surface texture was confirmed. The swelling was non tender on palpation, hard in consistency, non compressible, non reducible and non fluctuant. No bleeding or pus discharge was noted on palpation.

Intraorally, there was no swelling and the area was non-tender. The lower left third molar was fully erupted and the lower right third molar was partially erupted.

RADIOGRAPHICAL FINDINGS

Panoramic radiography showed a large oval unilocular radiolucency with sclerotic border. The size of the lesion was approximately 4 x 3 cm occupying the left mandibular ramus from the distal aspect of third molar tooth to the neck of condylar process and the coronoid process including the left ascending ramus area. No resorption of root was observed in the third molar. (fig.2)

Based on clinical and radiographic findings, provisional diagnosis of odontogenic tumour was made.

Odontogenic keratocyst and central hemangioma, aneurysmal bone cyst were considered in the differential diagnosis.

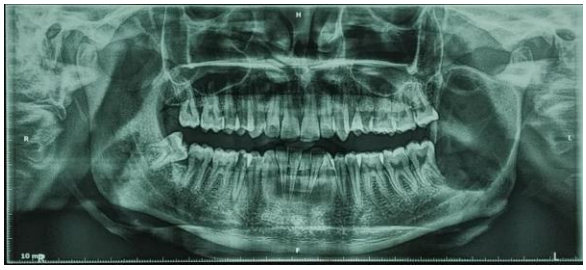

Fig.2

HISTOPATHOLOGICAL FINDINGS:

Fine needle aspiration cytology (FNAC) of the lesion was performed, which yielded straw- colored fluid. (fig.3)

Fig.3

FNAC report revealed presence of pus cells 2- 3/H.P.M.

Subsequently, the patient underwent incisional biopsy of the lesion.

H&E stained sections showed connective tissue stroma consisting of epithelium arranged in the form of anastomosing strands. The epithelial strands had outer tall columnar ameloblasts like cells and central stellate reticulum like cells. There was also presence of collagen fibers, numerous blood capillaries with extravasated RBCs, and inflammatory cells within the connective tissue stroma. The features were suggestive of plexiform ameloblastoma.

Based on clinicopathologic correlation, the diagnosis was confirmed as unicystic ameloblastoma of plexiform variant.

DISCUSSION

Ameloblastoma is a locally benign invasive tumor that has a high tendency to recur, metastasize and even undergo malignant transformation. It has a high tendency to recur, metastasize and even undergo malignant transformation. It has a high recurrence rate if not adequately removed[6] but local recurrence may occur even in patients who have undergone satisfactory primary surgical treatment.[7] As these tumors recur they become more aggressive and can develop into a lesion that is more aggressive than a sarcoma.[8] Recurrence seems to depend on several factors, such as (1) method of treatment of the primary lesion, (2) the extent of the lesion and (3) the site of origin.[9]

The radiographic appearance of an ameloblastoma varies from characteristic soap bubble loculations, to unicystic and multicystic radiolucencies, to subtle appearances such as expanded follicles of erupting teeth.[10]

The most common location is the posterior mandible associated with impacted teeth and follicular cysts, causing expansion of the cortical plates with scalloped margins and perforations with resorption of the involved teeth in advanced stages.[10,11]

Radiographically an ameloblastoma may be mistaken for a dentigerous cyst, aneurysmal bone cyst, central hemangioma or other central bony tumors.[11]

The 2005 WHO classification for ameloblastomas includes four subtypes. The solid/multicystic is the most common type, comprising 91 % of the ameloblastomas in the largest series.[12] This is followed by the unicystic type 6 %, the extra osseous ameloblastoma 2 %, and the desmoplastic type 1 %. The most aggressive clinical/pathologic association is seen in the solid/multicystic type, which is associated with the highest recurrence rate of up to 90 % with conservative operations such as enucleation and curettage.[13,14]

The unicystic type is the most benign and is further classified into luminal, intraluminal and mural subtypes. The luminal unicystic subtype does not exhibit invasion of the supporting connective tissue, has the lower recurrence rate of the three subtypes, and may be the only histology amenable to conservative surgical treatment.[13, 15-18]

CONCLUSION:

The diagnosis of unicystic Ameloblastoma was based on clinical, radiological and histopathological features. Around 52-100% of unicystic ameloblastoma are associated with impacted tooth (non dentigerous variant). This case report was an effort to bring forth a case of a large unicystic ameloblastoma which is not associated with impacted tooth.

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