ORIGINAL RESEARCH PAPER

Dental Science

ACANTHOMATOUS AMELOBLASTOMA- A RARE VARIANT IN A YOUNG FEMALE

KEY WORDS:

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ABSTRACT

Acanthomatous ameloblastoma histologically presents with squamous epithelial metaplastic transformation of odontogenic tissue. Because of unilocular presentation, it is commonly misdiagnosed as an odontogenic cyst. The present case report of a 25 years old female exclusively elaborates the issues concerned with the aggressive nature of acanthomatous ameloblastoma which is a distinct variant of ameloblastoma.

INTRODUCTION

Ameloblastoma is a benign but locally aggressive tumor of odontogenic origin. However the tumor cell do not form any enamel or hard tissue. It is the most frequent odontogenic tumor of the mandible and maxilla. Ameloblastoma can occur in all age groups but the peak incidence is seen in the 3rd and 4th decades of life with a rare occurrence in childhood. It is generally asymptomatic and presents as a slowly enlarging facial swelling. Radiographically, it appears either as unilocular radiolucent area with a well-defined margin or as a multilocular radiolucency with honeycomb appearence.[4] Ameloblastoma is infamous for its invasive growth and tendency to recur. According to the World Health Organization (WHO) classification of odontogenic tumors, ameloblastoma are divided into four types: Solid/Multicystic, Extraosseous/Peripheral, Desmoplastic and unicystic. This classification may have a prognostic value. Based on histopathology, ameloblastoma is classified into: Follicular, plexiform, acanthomatous, granular, basal cell, and desmoplastic. They can be found combined or isolated and are not related to prognosis of the tumor. The most prevalent histological subtype is the follicular variant (64.9%), followed by the plexiform (13%) and the acanthomatous variants (3.9%).

According to Larsonn and Almeren, incidence of the acanthomatous type has been 0.6 cases per million, whereas Shear and Singh found an incidence of 0.31 cases per million.Acanthomatous ameloblastoma mostly occurs in older patients compared to younger ones. Occurrence of tumor is more commonly seen in mandible than maxilla. The differential diagnosis prior to definitive treatment is crucial and necessitates a tissue biopsy as the lesion requires to be treated more aggressively than other benign lesions. This case report presents a case of a unicystic acanthomatous ameloblastoma in a 25 years old female.

CASE REPORT

A 25 year old female patient reported to the Department of Dentistry in G.G.S Medical College & Hospital, Faridkot with chief complaint of swelling of right side of lower jaw since one year. The swelling was initially small in size but gradually increased to present size (**Fig 1**). It was initially painless but she started experiencing intermittent pain occasionally. Medical history was non-contributory. The patient had visited various dentists previously and antibiotics/analgesic were prescribed but with no relief. On extra-oral examination, a diffuse swelling with asymmetry of face was present on right side. There was neither change in coiour of overlying skin nor any rise in temperature. The swelling was hard in consistency and non-tender in nature. There was no lymphadenopathy in head and neck region.

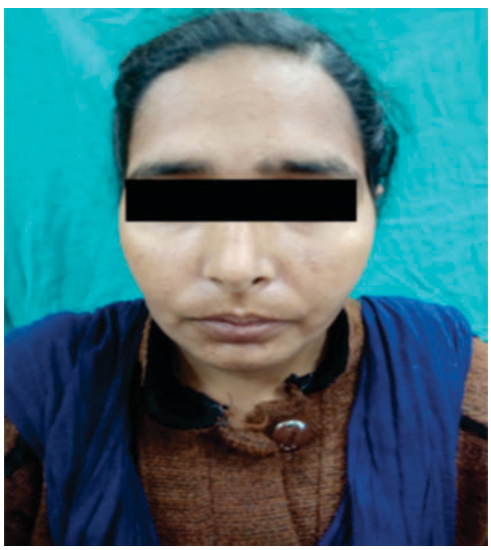
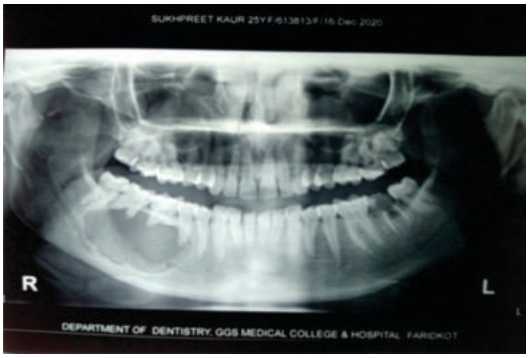


Fig 1: Clinical Photograph Showing Swelling On Left Side Of Face

Intraoral examination revealed that the swelling extended from distal of 44 to 48. There was no nerve deficit. Pre-operative OPG (Orthopantogram), (**Fig 2**) showed radiolucency extending from 44to 48.

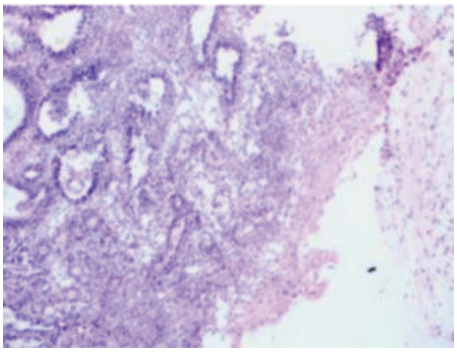
Fig 2: showing large Unilateral radiolucency on right side extending from 2nd premolar to 2nd molar



Radiolucency was unilocular, well demarcated with corticated margin. On aspiration, there was golden yellowish fluid resembling the cystic pathology. Routine hemogram was performed and all the blood investigations were within normal limits. Incisional biopsy was done under local anaesthesia and was sent for histopathological examination.

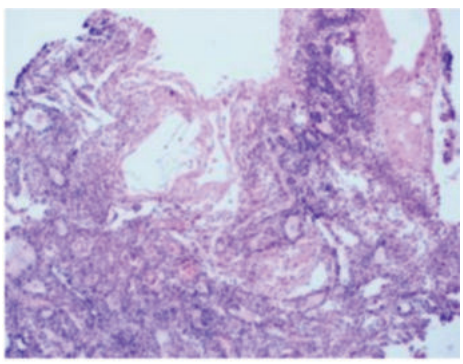
Section showed fragments of tissue containing tumour composed of anastomosing cords and larger sheets of odontogenic epithelium (**Fig 3a**).

Fig 3A: H& E Section Showing Sheets And Cords Of Odontogenic Epithelium Exhibiting Peripheral Palisading With Reverse Polarization(100X)



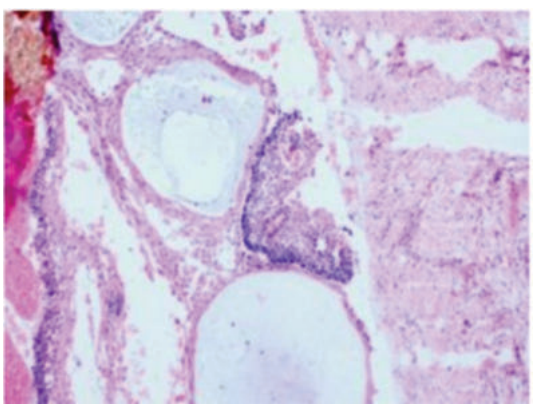
The cords and sheets of epithelium were bounded by columnar ameloblastic cells which were surrounding more loosely arranged epithelial cells. There were many areas in the centre of these showing squamous metaplasia . (**Fig 3b**).

FIG 3B: H& E Section Showing FOCI Of Squamous Metaplasia (100X)



There were many foci of microyst formation also noted (**Fig 3c**).

FIG 3C: H& E SECTION SHOWING FIBROUS CONNECTIVE TISSUE FORMATION ALONG WITH MICROCYST FORMATION (100X)



Biopsy report confirmed the diagnosis of acanthomatous amelobastoma. The patient was advised resection with reconstruction plate citing the chance of reoccurrence but patient denied the radical treatment and opted for conservative management.

Patient was scheduled for enucleation and curettage with extraction of four adjacent teeth. The surgical wound was then treated with Carnoy's solution to decrease the chance of reoccurrence after conservative treatment of ameloblastoma. Layerwise closure of wound was done. Suture removal was done on 10th day. Patient was recalled after one month for evaluation. No fresh complaints were reported by the patient .Patient was recalled after two months but she did not turn up for follow up afterwards.

DISCUSSION

Ameloblastoma is a benign but aggressive neoplasm of odontogenic epithelium, however, no enamel or hard tissue is formed by the tumor cells.[12] It comprises 1% of all radiolucent jaw lesions.[13] Ameloblastomas arise from either neoplastic transformation of odontogenic cyst epithelium or from residual epithelial rests left over from the formation of teeth, such as remnants of the enamel organ found over the crown of an unerupted tooth, rests cell of Malassez in the periodontal ligament or remnants of the dental lamina (rests of Serres). Ameloblastomas represent approximately 11 to 18% of all odontogenic tumors and are only second to odontomas. The prevalence seems to vary with the site of study as many hospital studies have reported higher prevalence than those of universities. Similar finding was observed at G.G.S Medical College & Hospital, Faridkot in past 11 years.

Though there is no gender predilection in the literature, a few studies have reported predominance in men. The reported ratio of mandibular to maxillary unicystic ameloblastoma is 13:1. Acanthomatous ameloblastoma is seen mostly in old age, the mean age being 60-62 years with a peak incidence in the 7 th decade of life.However, in our case, the patient was young female and was in third decade of her life.

The signs and symptoms of swelling and pain in our patient were similar to those reported in literature. Slow growth, being progressive and expansile are important behavioral characteristics of ameloblastoma., Consequently patient often presents with asymptomatic facial asymmetries at more advancing stages of the lesion.

The location of tumour shows marked predilection for mandible irrespective of its variant. The acanthomatous ameloblastoma is also found in mandible (81%) when compared to maxilla (19%).The posterior mandible is the most affected area. In our case too, the mandibular posterior region was involved.

Different modalities of imaging exams can be used for the evaluation, elaboration of the lesion and planning for the treatment of these tumors. These include periapical, occlusal, and panoramic radiograph, Computed Tomography (CT), Cone Beam Computed Tomography (CBCT) and Magnetic Resonance Imaging (MRI). CT and CBCT are more commonly used to evaluate the bone characteristics of the tumor, whereas MRI provides details about the soft tissues involved by the lesion. We have observed that OPG & CT scan are sufficient to access the lesions.

Radiographically, ameloblastomas are observed as unilocular or multilocular radiolucent lesions with well-defined borders, which can cause root resorption and displacement of the teeth involved by the lesion. In our case, OPG showed unilocular lesion with root resorption of the involved teeth and disruption of buccal and lingual cortical plates. Although the final diagnosis should be confirmed by histological examination, Differential diagnosis of

ameloblastoma includes odontogenic keratocyst, dentigerous cyst, calcifying odontogenic cyst, calcifying odontogenic tumor and ameloblastic fibroma.

Histopathological features of acanthomatous ameloblastoma includes all the general features of ameloblastoma such as connective tissue stroma having epithelial cell nests lined by tall columnar ameloblast like cells with reverse polarity. Stellate reticulum like polygonal cells in the central portion of the follicles may show cystic degeneration. Squammous metaplasia is a peculiar feature of acanthomatous ameloblastoma as was observed in our case along with microcyst formation.

Some authors suggest that the untreated acanthomatous variant can develop into an invading and metastasizing squamous cell carcinoma. Even, the biological behavior of the acanthomatous ameloblastoma is controversial. This may be because this variant is locally aggressive and frequently invades the alveolar bone or recurs after marginal surgical excision. Others suggest that there is no behavioral difference between the various subtypes of ameloblastoma and there is no special extensive local infiltration or bone destruction or risk of recurrence. There is no consensus on the pathogenesis and the invasive growth of ameloblastoma. This underscores the importance of, molecular mechanisms of cell proliferation. To predict aggressiveness of ameloblastoma, particularly which is a P16 cyclin-dependent kinase inhibitor and is a tumoral suppressor protein encoded by the *CDKN2A* gene. The immunosuppression of the P16 protein may be inversely related to aggressiveness and rate of reoccurrence.

The recommended treatment for ameloblastoma is radical resection of jaw. However the decision should be based on variant, location, clinical behaviour and size of the tumor as well as age of the patient. As radical resection affects facial esthetics and maxilla-mandibular relationships, conservative surgical treatment should be considered whenever the situation demands.

Among the various types of ameloblastomas, multicystic ameloblastoma have a much higher rate of recurrence than unicystic ameloblastoma. The reason for this higher rate is believed to be because of the numerous microextensions that the tumor has projecting into the bone. The recurrence seems to depend on several factors such as method of treatment of primary lesion, extent of the lesion and site of lesion.

CONCLUSION

Among all the variants of ameloblastoma, unicystic ameloblastoma is the least common and acanthomatous variant is even rare. The management involves meticulous approach for differential diagnosis and thorough investigations. The treatment options consist of conservative and radical resection of jaw. In conservative management, enucleation followed by application of Carnoy's solution is the standard procedure to prevent recurrence. At the same time radical treatment reduces the chance of occurrence also.

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