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CASE REPORT

Ameloblastoma of the Mandible: A Case Report with Review of Literature

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ABSTRACT

The ameloblastoma, particularly the mixed cystic/solid type, is the most clinically significant odontogenic tumor. The tumor is often locally aggressive and has a significant impact and may have a patient's morbidity and mortality. In this report, we present a case of a large ameloblastoma that presented with the typical radiographic features of variably sized radiolucent loculations. Microscopically, the tumor showed a pattern of acanthomatous variant predominating. The tumor was treated with a partial resection of the mandible.

Keywords: Acanthomatous ameloblastoma, Squamous metaplasia, Mixed cystic/solid type.

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INTRODUCTION

Ameloblastomas are an enigmatic group of oral tumors whose importance lies in its potential to grow into enormous size with resulting bone deformity.[1] Their name implies a resemblance to cells of the enamel-forming organ. It represents about 1% of all oral ectodermal tumors and 9% of odontogenic tumors. It is an aggressive neoplasm that arises from remnants of the dental lamina and dental organ (odontogenic epithelium).[2] They are typically classified as multicystic, unicystic, peripheral and malignant subtypes.[1] 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 and keratoameloblastoma.[3] This report illustrates a case of potential complication of dentigerous cyst into ameloblastomatous variant of acanthomatous type of the mandible in an 46-year-old female.

CASE REPORT

A 46-year-old female reported to the Department of Oral Pathology and Microbiology, Indira Gandhi Government Dental College, Jammu, with a complaint of swelling over the right side of face for past 2 years showing facial deformity. On extraoral examination, a large painless swelling was seen which was hard in consistency and nontender about 3 × 3 cm in diameter showing facial

Fig. 1: Swelling present over the right side of the mandible

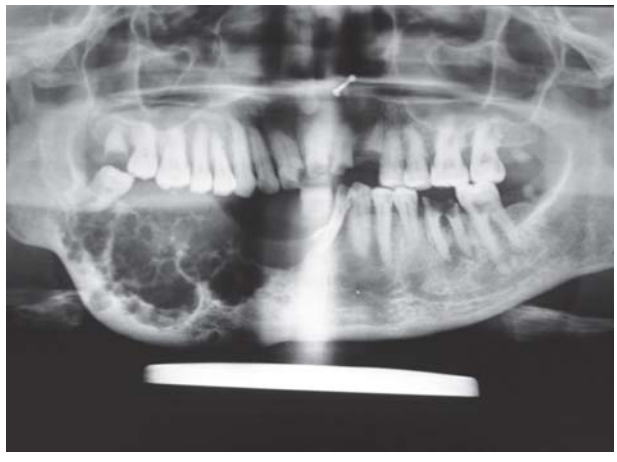

Fig. 2: Multilocular radiolucency extending from the ramus region to the angle and body of mandible associated with partial destruction of the lower border of mandible

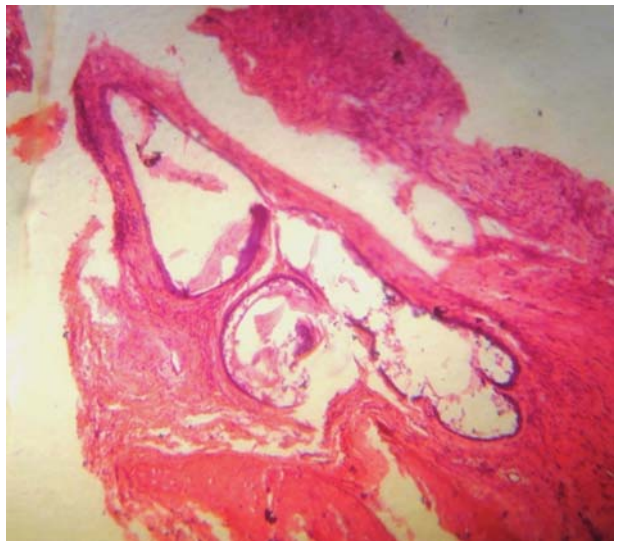

Fig. 3: Thin epithelial lining of one to two cells layer thick (10×)

asymmetry. Intraoral examination revealed very large swelling extending from lower right canine to the angle of the mandible (Fig. 1).

OPG revealed a large multilocular radiolucency extending from the ramus region to the angle of mandible associated with partial destruction of the lower border of mandible (Fig. 2).

A clinical diagnosis of ameloblastoma/odontogenic keratocyst was made. Excisional biopsy was done and specimen submitted for histopathological examination. Microscopically, hematoxylin and eosin-stained section showed the presence of thin epithelial lining of one to two cells layer thick (Fig. 3). In the underlying connective tissue, follicle of epithelial islands were seen showed columnar cells which appear like ameloblast and in the center stellate reticulum like cells were seen (Fig. 4). These follicles also showed the presence of squamous metaplasia at few places

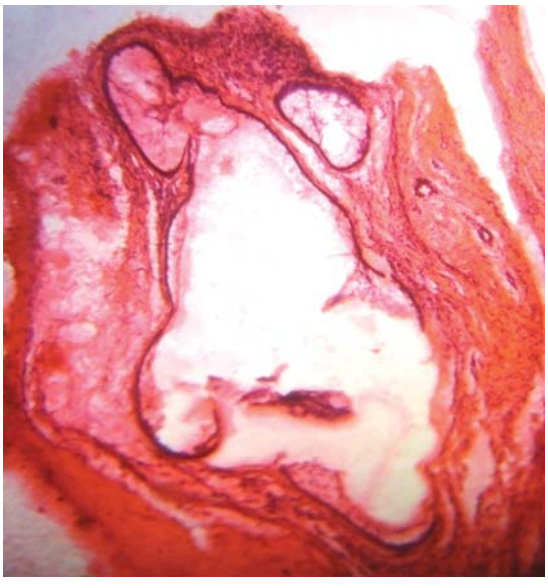

Fig. 4: Connective tissue stroma showing follicle of epithelial islands with stellate reticulum like tissue in the center (4×)

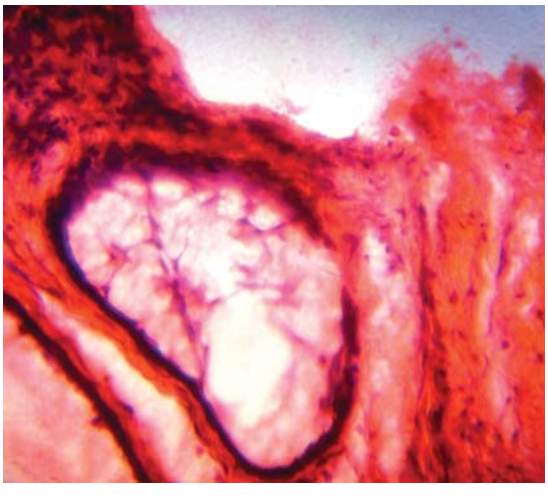

Fig. 5: Follicles with squamous metaplasia were also seen (40×)

supported by fibrous connective tissue (Fig. 5). Few areas of hemorrhage and blood vessels were also evident. Based on the above features, a diagnosis of dentigerous cyst undergoing ameloblastomatous transformation of acanthomatous variant was made.

DISCUSSION

Gorlin identifies Cusack as the first person to identify ameloblastoma in 1827. Falkson gave a detailed description in 1879. The first histopathologic description was given by Wedl (1853) who called the tumor cystosarcoma or cystosarcoma adenoids and thought that it could have arisen from tooth bud/dental lamina. Wagstaffe (1871) gave the first histological drawing. Malassez (1885) introduced the term 'adamantine epithelioma', while Derjinsky (1890) introduced the term 'adamantinoma'. However, this term has become obsolete and has to be avoided. Ivy and Churchill in 1930 encouraged the use of the term 'ameloblastoma' which is the preferred terminology till date.[4]

Ameloblastoma, although rare, is the most common odontogenic tumor accounting for 1% of all tumors in head and neck region and around 11% of all odontogenic tumors.[5] In the newest classification by the World Health Organization, variants of ameloblastoma are categorized on the basis of characteristics, such as the age at presentation, location in the body, imaging features, clinical behavior, and prognosis. Thus, the plural term ameloblastomas is used to describe this family of diseases.[6] Ameloblastomas are classified as either extraosseous (peripheral) or intraosseous. Peripheral ameloblastomas manifest as a sessile or pedunculated slow-growing mass that is confined to the gingiva or alveolar mucosa with no involvement of underlying bone. Intraosseous ameloblastomas arise in the jaw and are further classified as unicystic, desmoplastic and mixed cystic and solid.[7] The mixed cystic and solid form demonstrates more aggressive behavior and is more likely to recur than unicystic and desmoplastic ameloblastomas.[7] Larsson and Almeren report the incidence of ameloblastoma in Sweden as 0.3 cases per million people per year. The average age of patients with intraosseous ameloblastoma has been reported to be 39 years and appears equal frequency between sexes, although a higher frequency in females than in males has been described.[6-8] In the present case, the patient was female and was in fourth decade of her life. Similar to the findings of Fregnari et al[9], 80% of our tumors were located in the mandible, 70% are located in the area of the molars or the ascending ramus, 20% in the premolar region and 10% in the anterior region.[5] About 10 to 15% of ameloblastomas are associated with a nonerupted tooth.[2] In

the present case, a large acanthomatous ameloblastoma found in the ascending ramus and molar region of the mandible and it was associated with a erupted tooth. Clinically, it frequently manifests as a painless swelling, which can be accompanied by facial deformity, malocclusion and paresthesia of the affected area. In our case, clinical examination revealed a large, expansive mass in the ascending ramus and molar region of the mandible. The swelling was hard, painless to palpation and covered by normal mucosa.

At radiography,[10] the mixed cystic and solid type of ameloblastoma appears as an expansile, radiolucent, uni- or multilocular mass with internal septations that form a honeycomb or soap bubble appearance at all modalities, a classic finding which was also demonstrated in the present case.

Ameloblastomas may arise from various sources of odontogenic epithelium, including the epithelial lining of the dental follicle. Approximately, 50% of ameloblastomas arise from the epithelial lining of a dentigerous cyst.[11] In the present case, the association with presence of non-specific thin epithelium lining in focal areas of cystic tumor supporting the second hypothesis, i.e. arising from preexisting dentigerous cyst.

Optimal treatment of ameloblastomas consists of wide surgical resection. Radiation therapy may be considered, if complete resection is not possible or if positive resection margins are not amenable to resection.[11] Dentigerous cysts usually are treated with enucleation and curettage, whereas the preferred treatment of keratocystic odontogenic tumors is enucleation with wide bone margins and marsupialization because of their aggressive growth, multiplicity and likelihood of recurrence with more conservative treatment.[12] A follow-up of ameloblastoma is necessary because 50% of all recurrences are within 5 years postoperatively.[1]

CONCLUSION

Our case is consistent with three entities: An erupted third molar tooth, a mixed cystic and solid ameloblastoma and a dentigerous cyst. The coexistence of these three entities also is in keeping with the hypothesis of ameloblastic transformation of dentigerous cysts.

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