CASE REPORT

Large Plexiform Ameloblastoma of Anterior Mandible: A Case Report and Review of Literature

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Introduction

Ameloblastoma is an epithelial odontogenic neoplasm mostly of enamel organ-type tissue that has not undergone differentiation to the point of hard tissue formation[1]. It represents 10% of all the tumors of the jaw bone[2]. Ameloblastomas are benign but locally invasive neoplasm and a low propensity to metastasize[3]. These arise in the molar-ramus area of the mandible, and are occasionally associated with unerupted third molar teeth[4].

Ameloblastoma appears most commonly in the third to fifth decades but the lesion can be found in any age group including children[5].They are usually asymptomatic, usually recognized on routine radiographic examination but may be associated with jaw expansion ,root displacement, root resorption and facial disfigurement[4]. Ameloblastomas are reported to be more common in dark skinned people and in developing nations[6].

The chief histopathological variants of ameloblastoma are the follicular and plexiform types, followed by the acanthomatous and granular cell types and desmoplastic, basal cell, clear cell ameloblastoma, keratoameloblastoma and papilliferous ameloblastoma are uncommon variants [4].Radiologically they are unilocular or multilocular radiolucency with a honeycomb or soap bubble appearance.

Case Report

A 22-year-old Indian male presented with a swelling in anterior region of lower jaw for 5 months. Clinical examination revealed a diffuse swelling in the anterior mandible, measuring 6 cm x 4 cm in size, with obliteration of the labial and lingual vestibule (Figure 1).

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Figure-1: Intraoral view shows obliteration of labial and lingual

The swelling was non-tender. Mucosa over the swelling appeared ulcerated. There were no palpable lymph nodes in the cervical region. A review of other systems did not reveal any significant findings and haematological findings were within normal limits.

A panoramic radiograph revealed a multilocular radiolucency extending from 37 to 47 **(Figure 2).** Root resorption was seen in relation to 33, 34, 35, 36 and 47. The base of the mandible was damaged and thinned.

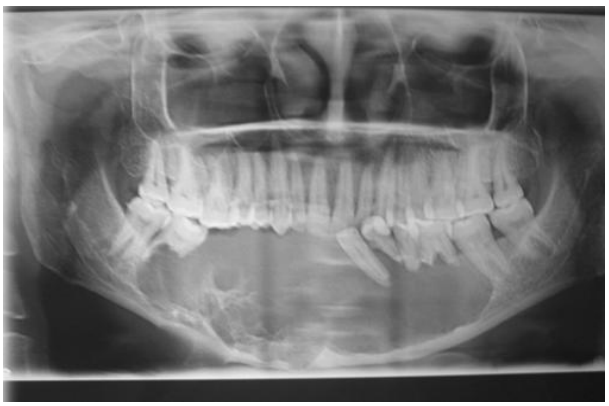

Figure-2: Radiograph showing multilocular radiolucency extending from 37 to 47 and root resorption in relation to 36, 35, 34, 33 and 47.

An incisional biopsy was done and the specimen was sent for histopathological examination.

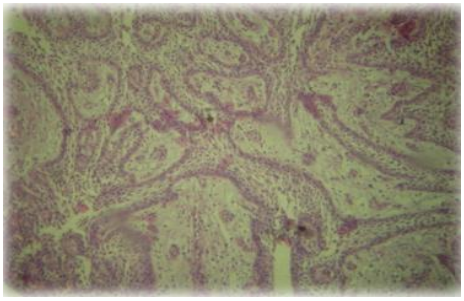

Figure 3: Photomicrograph demonstrates a plexiform ameloblastoma predominantly composed of the epithelium arranged as a tangled network of anastomosing strands.

Histopathology revealed odontogenic epithelium arranged as a tangled network of anastomosing strands with peripheral tall columnar cells exhibiting reversal of polarity resembling ameloblasts. The central cells were loosely arranged resembling stellate reticulum with areas of extensive cystic degeneration. The supporting connective tissue stroma showed moderate vascularity, moderate chronic inflammatory cell infiltrate. The histopathological diagnosis was plexiform ameloblastoma **(Figure 3).** Under general anaesthesia a segmental resection of anterior mandible was performed. The histopathology of the excisional biopsy specimen was consistent with the findings of the incisional biopsy report. Two years later the patient underwent microvascular reconstruction of the mandible.

Discussion

Generally, odontogenic tumours have been reported to be rare and accounts for only 1 % of the jaw tumors [4]. Numerous histological patterns have been described in ameloblastomas and most common being follicular and plexiform types. The most common location of ameloblastomas is posterior regions of jaw, but this case is a rare case of plexiform ameloblastoma involving anterior mandible.

Ameloblastoma is a benign locally invasive neoplasm with high rate of recurrence. Shafer et al. postulated that ameloblastomas arise from either cell rests of the enamel organ,epithelium of odontogenic cysts, disturbances of the developing enamel organ,basal cells of the surface epithelium or heterotropic epithelium in other parts of the body[7].

Ameloblastoma can appear at any age but 3rd to 5th decade is most commom and does not show any predilection towards any gender, but in our case the patient is 22 years of age. The most favored site is the ascending ramus (70%) followed by the premolar region (20%), anterior region (10%) and 10-15% are associated with a non-erupted tooth[4].Clinically, it usually manifests as a painless swelling, and can be accompanied by facial deformity, malocclusion, ulceration, periodontal disease and paresthesia of the affected area[8].

Kim et al. stated that ameloblastoma is characterized by the proliferation of epithelial cells arranged on a stroma of conjunctive vascular tissue in locally invading structures that resemble the enamel organ at different stages of differentiation. Diverse histological patterns have been described in the literature and include follicular, plexiform, acanthomatous, papilliferous-keratotic, desmoplastic, granular, vascular and those with dentinoid induction. The tumor found in our patient was an ameloblastoma of the plexiform type. The term plexiform refers to the appearance of anastomosing islands of odontogenic epithelium in contrast to a follicular pattern[9].

Histopathological features in our case showed anastomosing sheets and cords of odontogenic epithelium. The epithelium displayed tangled network of anastomosing strands, a stellate, reticulum-type appearance, arranged as enclosing cysts of various sizes.

Conclusion

Ameloblastomas are enigmatic group of oral neoplasms that can occur not only in posterior region but also in anterior region of jaw. Thorough knowledge of the ameloblastic neoplasms is significant to clinician for effective treatment strategies and to prevent recurrences.

References

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