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Case Report

Unicystic Plexiform Ameloblastoma of mandible-A rare case entity

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Abstract

Ameloblastoma is the most common among the epithelial odontogenic tumors, but it is still comparatively rare, comprising approximately 1% of tumors and cysts arising from the jaw. It appears most commonly in the third to fifth decades, but it has also been described in children. No gender or racial preference has been noted. More than 80% occur in the mandible, with 70% of these arising in the molar–ramus area.

Keywords: Unicystic Plexiform Ameloblastoma, plexiform pattern tumors, mandible tumors.

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INTRODUCTION

Ameloblastoma is the most common among the epithelial odontogenic tumors, but it is still comparatively rare, comprising approximately 1% of tumors and cysts arising in the jaws. It appears most commonly in the third to fifth decades, but it has also been described in children. No gender or racial preference has been noted. More than 80% occur in the mandible, with 70% of these arising in the molar–ramus area. The usual radiographic appearance is that of a lytic expansile lesion. Clinical duration may range from a few weeks to 50 years.

The plexiform pattern demonstrates irregular masses and interdigitating cords of epithelial cells. The central portion of the epithelial island is composed of a loose network of cells resembling stellate reticulum.

Case Report

A 52-year-old female presented with pain over left lower gums and teeth since 1 year. Radiological examination, showed a large unilocular radiolucent lesion on the anterior aspect of left ramus of mandible. She underwent segmental mandible resection from 35 to 38 region (lower left-quadrant III). We received a tumor of size 2.2×2×0.9 cm involving the mandible and anterior aspect. External surface was tan brown with congested blood vessels. Cut surface was cystic filled with brownish material.

On microscopic examination showed bony trabeculae and marrow infiltered by a neoplasm composed of sheets, irregular nests and anastomosing strands of squamous cells with moderate eosinophilic cytoplasm and uniform to mildly pleomorphic vesicular nuclei (Fig.1&Fig.2). The strands are separated by fibrous septae with numerous congested and dilated blood vessels (Fig.4). Focal areas of necrosis seen. The resected margins showed no tumor infiltration.

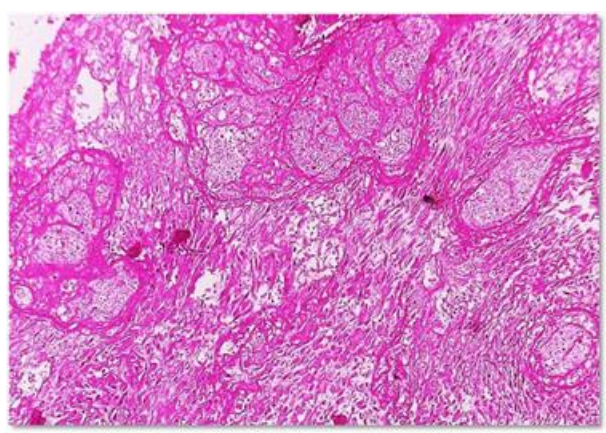

Fig.1: Histopathological section showing neoplasm composed of peripheral palisading of epithelial cells and stellate reticulum-like areas. (10X**)**

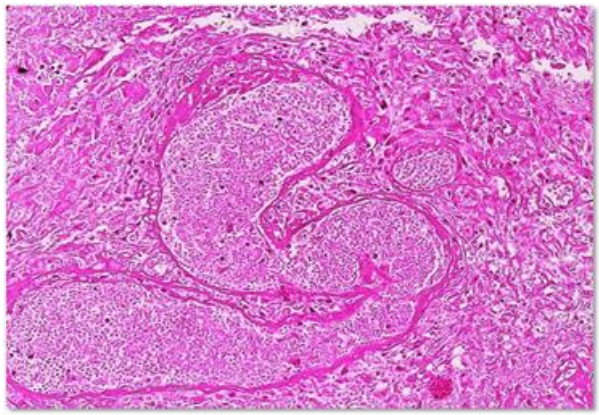

Fig.2: Neoplasm composed of irregular nests and anastomosing strands of squamous cells with moderate eosinophilic cytoplasm and uniform to mildly pleomorphic vesicular nuclei.

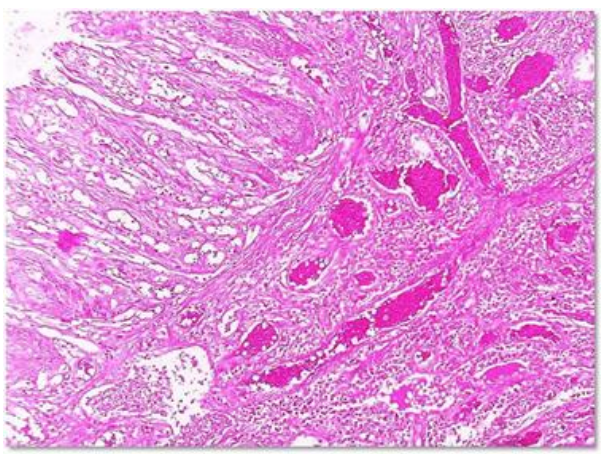

Fig.3: The strands are separated by fibrous septae with numerous congested and dilated blood vessels.

The term Plexiform Ameloblastoma refers to a pattern of epithelial proliferation arising from cystic lesion of the jaws.[1] It does not exhibit the histological criteria for ameloblastoma published by Vickers and Gorlin, and has therefore been considered by some pathologists to be a hyperplastic epithelial proliferation of the cystic lining rather than true ameloblastoma. [2] Plexiform ameloblastoma present with typical features of cords and sheets of anastomosing odontogenic epithelial cells and might show features of amelobastoma, such as peripheral palisading, reverse polarity of basal cells, stellate reticulum-like areas. Treatment modalities for Unicystic Plexiform Ameloblastoma have been used such as enucleation, followed by application of Carnoy's solution, marsupialization followed by surgery, and segmental resection. The recurrence rate after enucleation alone is highest 30.5%, while resection of this tumor results in the lowest recurrence rate 3.6%(6,7,8).

In Sivapathasundharam and Einstein study [3], patient presented with diffuse non tender, smooth surfaced hard swelling over left posterior aspect of jaw. On radiology unlike our case, showed a large multilocular radiolucency extending from 1st premolar to angle of mandible. On histopathological examination, luminal and intramural plexiform epithelial proliferation with typical dentin in connective tissue capsule.

In Yavagal et al study[4], patient presented with painful swelling, fluctuant in some areas and hard in some cases, extending from medial surface of 34-distal margin of 36. On radiology, well defined radiolucency from periapical region of 35–36 to inferior border of mandible. On histopathological examination, luminal plexiform pattern with subepithelial hyalinization and odontoblastic rests were seen within fibrous capsule.

Conclusion

The main aim of reporting this case was to discuss the clinical features, radiographic findings and histological presentation in plexiform ameloblastoma. According to recent literature these lesions are more aggressive than previously thought. Most of the times these lesions are misdiagnosed as a dentigerous cyst both clinically as well as radiographically. Hence, its essential for proper histopathological examination in evaluating plexiform ameloblastoma due to bad prognosis and aggressive behavior.

Conflict of Interest

There is no conflict of interest.

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Nil

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published.

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