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Original Research Paper

Oral Pathology

AMELOBLASTOMA WITH MULTIVARIANT HISTOLOGIC PATTERNS- A RARE CASE REPORT HIGHLIGHTING REVIEW OF LITERATURE

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ABSTRACT Ameloblastoma is a benign but locally aggressive odontogenic tumor accounting for 9-11% of all odontogenic tumors. It is the most common odontogenic neoplasm in India Here we report a rare case of ameloblastoma in a 37 years old male patient presenting with multiple histologic patterns along with a brief review of literature.

KEYWORDS : Ameloblastoma, histologic patterns, granular cell, desmoplastic

INTRODUCTION

Ameloblastoma is a benign but locally aggressive odontogenic tumor accounting for 9-11% of all odontogenic tumors. It is a true neoplasm of enamel organ type tissue, which does not undergo differentiation to the point of enamel formation. It is the second most common odontogenic neoplasm with odontoma outnumbering it in reported frequency of occurrence and is the most common odontogenic neoplasm in India.(Sivapathasundharam, 2020)

Ameloblastoma is commonly seen in young adults, with a mean age of 35 years and w no gender predilection. 80% of cases occur in the mandible, with molar, angle, ramus region most commonly affected.(Masthan et al., 2015) According to the 2022 WHO classification of odontogenic and maxillofacial bone tumours, Ameloblastoma is classified into unicystic, conventional, peripheral, metastasizing, and adenoid ameloblastoma which was newly added. A malignant counterpart also exists; the ameloblastic carcinoma. The conventional ameloblastoma includes six histological subtypes; follicular, plexiform, acanthomatous, granular, desmoplastic and basaloid.(Vered & Wright, 2022) Usually ameloblastomas show predominance of a single histologic subtype. Rarely it can show more than one histologic pattern. But to the best of our knowledge, this is the first case of ameloblastoma showing five different histologic patterns. Hence this case is being reported highlighting the literature.

CASE REPORT

A 37-year-old male patient reported to our institution complaining of a painless swelling in the lower left back tooth region since 2 months. History revealed extraction of mobile lower left third molar which was partially erupted 2 months prior. A few days after the extraction, the patient noticed a small swelling in the extraction socket which gradually increased in size.

Extra oral examination revealed a diffuse swelling in left submandibular region extending from posterior border of the mandible to 3cm anteriorly and 1cm inferiorly.

Intra orally, a firm, non-tender, proliferative growth of approximate size 2 x 1.5cm was seen in the lower left posterior alveolus in third molar region.

Orthopantamogram, CBCT and CT revealed a multilocular radiolucent lesion in posterior region of mandible extending from distal aspect of second molar to the ramus causing significant bone destruction. Radiating spicules were noted from the lower border inferiorly. There was minimal expansion of buccal cortex, multiple areas of cortical break mandibular canal erosion and complete destruction of lingual cortex.

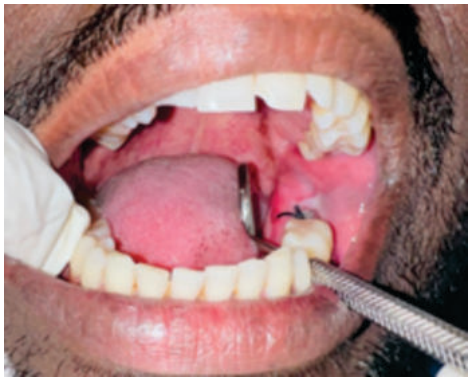

Figure 1- intraoral photograph

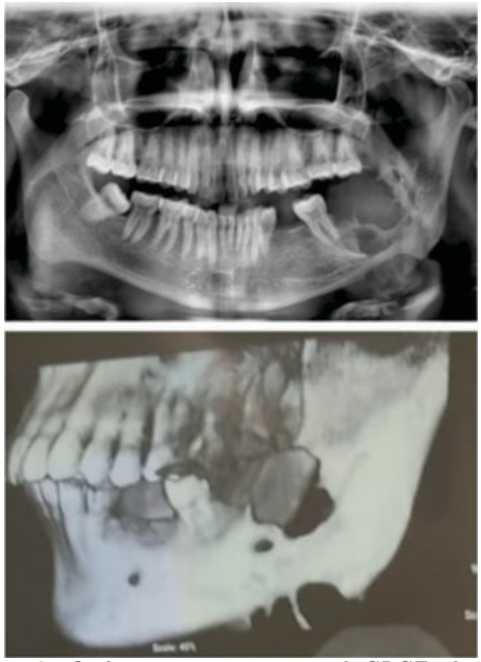

Figure 2- Orthopantomogram and CBCT showing multilocular radiolucent lesion in left posterior mandible

The radiographic impression was an aggressive lesion possibly Ameloblastoma. Routine blood investigations were done and were within normal limits.

An incision biopsy was done which on examination showed tall columnar cells proliferating as anastomosing chords with back-to-back cell arrangement showing palisaded basophilic nucleus with reversal of polarity and subnuclear vacuolation along with stellate reticulum like areas which led to the diagnosis of plexiform ameloblastoma. Hematoxylin and Eosin-stained section of excised tumor showed cuboidal to low columnar cells proliferating in plexiform pattern and follicles of tumor cells with central stellate reticulum like areas and peripheral cuboidal to low columnar cells showing hyperchromatic nuclei, reversal of polarity and subnuclear vacuolation. Perifollicular hyalinization was also seen. At areas, stellate reticulum like areas were replaced by large round cells with pale eosinophilic granular cytoplasm with centrally placed nucleus resembling granular cell pattern. Cytoplasm of peripheral cells were also showing granular cell changes. Acanthomatous pattern was also noted with extensive squamous metaplasia. Also, an area of increased collagen density with squeezed odontogenic epithelial islands resembling desmoplastic ameloblastoma.

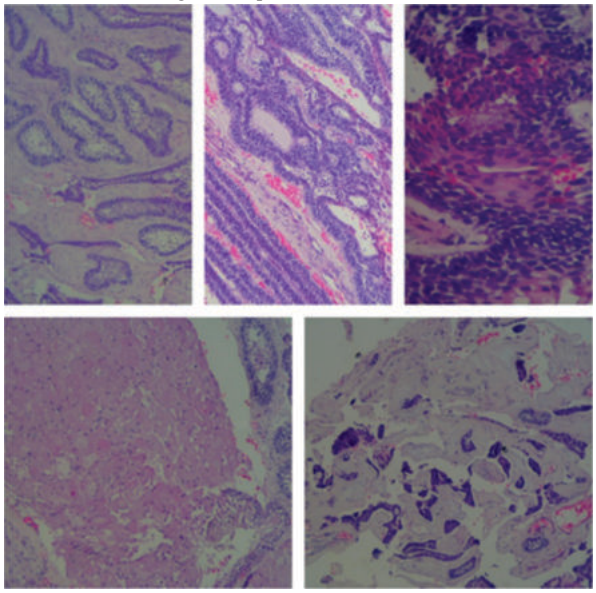

Figure 3- H&E stained section showing various histologic patterns- follicular, plexiform, acanthomatous, granular cell, desmoplastic

A definitive diagnosis of Ameloblastoma with plexiform, follicular, granular cell, acanthomatous and desmoplastic patterns was rendered. Post surgical period was uneventful and patient is under regular follow up.

DISCUSSION

A multitude of successive genetic and epigenetic processes culminate in odontogenesis. Disruptions in these processes can cause epithelial proliferations resulting in odontogenic tumours like Ameloblastoma where tumour cells arise from cell rests of serres and dental lamina, lining of odontogenic cysts, and basal cells of oral epithelium. Mutations in the genes of the mitogen-activated protein kinase (MAPK) pathway known to be active during odontogenesis, is a common occurrence in 90% cases of ameloblastoma. BRAF V600E and hedgehog-related SMO mutations are the most common mutation seen. RAS and FGFR2 are other mutations showed in MAPK pathway. Adenoid ameloblastoma is however an exception showing β-catenin mutation. (Sivapathasundharam, 2020; Soluk-Tekkesin & Wright, 2022; Vered & Wright, 2022)

Literature revealed ameloblastoma with combined histological patterns. Follicular and plexiform patterns occur concomitantly with mandible being the most frequent site.(Hertog et al., 2012) When desmoplastic ameloblastoma occurs in combination with other histologic patterns the term hybrid ameloblastoma is used. Kumar H et al(2016)(Kumar et al., n.d.) reported a case of ameloblastoma of the mandible in 27 yrs old male patient with follicular, plexiform and granular patterns. To the best of our knowledge this is the first case to be reported with co-occurrence of five different histologic patterns in intraosseous ameloblastoma except a case of peripheral ameloblastoma reported by Bajpai et al (2015)(Bajpai & Pardhe, 2015) in a 44 yrs old female showing five patterns.

The histologic subtypes may have prognostic implications for recurrence, follicular pattern is found to show the highest recurrence rate of 29.5% followed by plexiform 16.7% and the acanthomatous showing the least recurrence of 4.5%. But according to studies by Reichart et al (1995)(Reichart et al., 1995) recurrence rate of granular cell ameloblastoma was 33.3% exceeding the other types. However, the biologic behaviour of this tumour does not have any correlation with the histologic pattern and should be treated uniformly.

Granular cell change in ameloblastoma was first reported by Krompecher in 1918. Later in 1984 Nasu et al(Nasu et al., 1984) identified the cytoplasmic granules as lysosomal aggregates. Kumamoto et al (2001) suggests that granular cell change is due to increased apoptotic cell death and subsequent phagocytosis by neighbouring cells. Some others consider it a transitional stage of ameloblasts and occurs due to lysosomal insufficiency. Also dysfunction of lysosomal enzyme or enzyme related proteins have been suggested as the cause.(Yamunadevi et al., 2014)

Desmoplastic ameloblastoma is a also a rare variant of ameloblastoma with maxilla being the common site showing unique mixed radiolucency and radiopacity which according to Takata et al(1999) could either be due to prominent osteoplasia or due to peculiar infiltrative nature of the tumour to adjacent marrow spaces leaving the non-neoplastic bone to remain within the tumour.(Takata et al., 1999)

CONCLUSION.

Ameloblastoma appears with varied histologic patterns and often a mixture of patterns. Whether the mixed histologic type emerges from a tumour with single pattern or they originate as a mixed histologic pattern remains uncertain. However, the occurrence of five different patterns in a single specimen is extremely rare and more studies are required to identify the pathogenesis behind the occurrence of such multiform lesions and its prognostic significance.

Conflicts Of Interest

The authors declare that they have no conflict of interest.

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