UNICYSTIC AMELOBLASTOMA OF MANDIBLE –A DIAGNOSTIC DILEMMA

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Abstract

Unicystic ameloblastoma is a benign, cystic locally invasive neoplasm of odotogenic origin. Its clinical, radiographic and gross features are similar to the odontogenic cysts but histologically it shows ameloblastomatous epithelial lining with or without luminal or mural tumor proliferation. It is thought to be less aggressive than multicystic ameloblastoma. The article presents a case of large symptomatic unicystic ameloblastoma of mandibular molar region which was treated by enucleation under suspicion of residual cyst. The neoplastic nature of lesion became evident only after histological examination. The article also describes the importance and complexity of differential diagnosis of odontogenic lesion showing common clinical and radiographic features.

Key Words: Residual cyst, Mandibular molar region, Unicystic ameloblastoma

Introduction

Ameloblastoma as described by Robinson is a benign tumor that is "usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent".[1] It originates from cell rests of enamel organ, odontogenic rests of Malassez, reduced enamel epithelium and epithelium of odontogenic cysts.[2] Ameloblastomas are usually asymptomatic and found on routine dental radiographs; however, they may also present with jaw expansion. Radiographically, ameloblastomas can either be uni or multilocular with well-circumscribed margins. Ameloblastomas are typically differentiated into unicystic intraosseous, multicystic, solid intraosseous (80-90% of all ameloblastomas) and peripheral. The term unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth.[3] The unicystic ameloblastoma is a less encountered variant of the ameloblastoma. It appears more frequently in the second or third decade with no sexual or racial predilection.[4] It is almost exclusively encountered asymptomatically in the posterior mandible. Treatment for ameloblastomas ranges from curettage to en bloc excision depending on histology and clinical presentation of the tumor.[5] The present case is of symptomatic unicystic ameloblastoma of the mandibular molar region in a 16-year-old patient.

Case report

A 16-year-old female patient with non-contributory medical history reported for dental evaluation to outpatient department with chief complaint of a painful swelling in mandibular right posterior region since one month. Figure1.

Intraoral examination reveal a swelling associated with missing mandibular first molar which was carious and extracted, extending from first premolar to second molar antero-posteriorly and obliterating the buccal vestibule. The overlying mucosa was smooth and there was no colour change. On palpation, the swelling was tender, hard, with slight softness on buccal side. The panromic radiograph revealed a well-defined unilocular radiolucency in relation to missing permanent mandibular right first molar extending from distal surface of permanent first premolar to mesial surface of permanent second molar. Figure 2

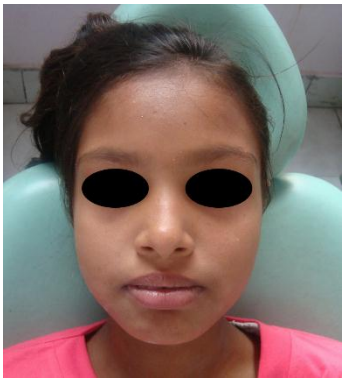

Figure 1: - Swelling on right side of face.

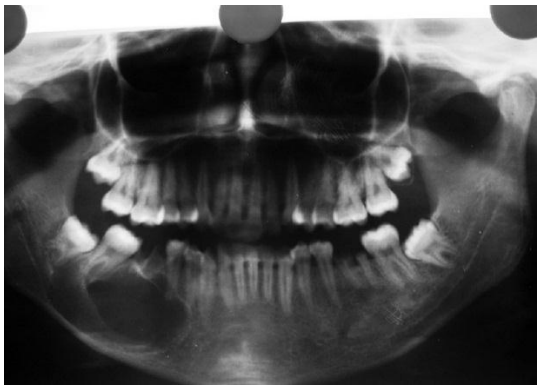

Figure 2: - Orthopentomogram showing uni-locular cystic lesion.

On basis of history, clinical and radiographic examination, provisional diagnosis of residual cyst was made. Enucleation of the lesion was performed under local anesthesia. Grossly the soft tissue specimen was greyish white in colour firm in consistency and 4 x 3 x 2 cm in dimensions. Figure 3

Histopathological analysis of the surgical specimen showed a cystic space surrounded by fibrous capsule. The cystic lumen was lined by epithelium of variable thickness consisting of ameloblast like cells fulfilling the Vickers and Gorlin criteria i.e. the basal cell layer composed of hyperchromatic tall columnar cell with palisaded nuclei

showing reverse polarity and subnuclear vacuolization. A thin overlying layer of stellate reticulum like cell were also evident. Figure 4 & 5

Figure 3: - Surgical Tissue specimen

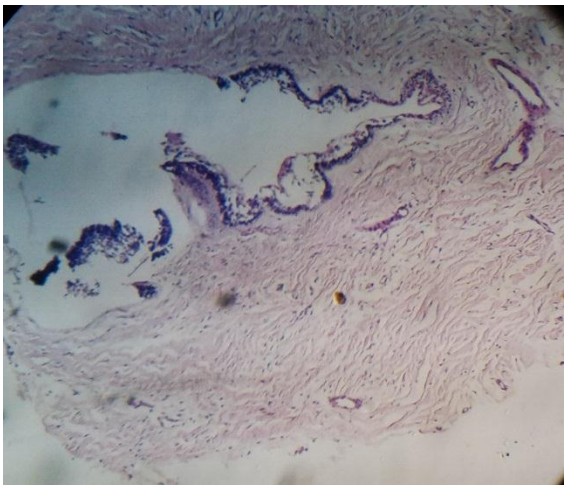

Figure 4: - Cystic lining surrounded by fibrous capsule (H&Ex10)

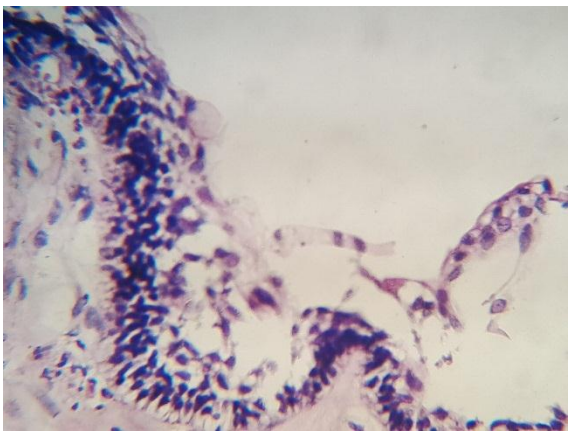

Figure 5: - Ameloblatomatous epithelium with overlying stellate reticulum like cells (H&Ex40)

Diagnosis of a simple unicystic ameloblastoma was made.

Discussion

Unicystic ameloblastoma, a variant of ameloblastoma first described by Robinson and Martinez in 1977. [3,6] This accounts for 10-15% of all intraosseous ameloblastoma.[7] Patients most commonly present with swelling and facial asymmetry, pain being an occasional presentation symptom. Small lesions are sometimes discovered more on routine radiographic screening or as a result of local effects like tooth mobility, occlusal alterations and failure of eruption of teeth, produced by the tumor.[8] Between 50 and 80% of cases are associated with tooth impaction, the mandibular third molar being most often involved.[9] In the present case swelling was present at the first molar region, symptomatic, firm with fluctuance noted on buccal surface which is not usual. In the most comprehensive study on the radiographic aspects of unicystic ameloblastoma, it was found that the unilocular: multilocular ratio was 13:3 for cases associated with impacted teeth.[10] Unicystic ameloblastoma is believed to be less aggressive, tends to affect patients at a younger age and its response to enucleation or curettage is more favourable than the classic solid or multicystic ameloblastomas. Based on the character and extent of tumor cell proliferation within the cyst wall, several histologic subtypes of unicystic ameloblastoma are recognized, which include those of simple cystic nature, those with intraluminal proliferative nodules and those containing infiltrative tumor islands in the cyst walls.[11] Other odontogenic lesions share common clinical and radiological features includes dentigerous cyst, residual cyst, odontogenic keratocyst, adenomatoid odontogenic tumor and giant cell lesions. Keratocyst usually spread anterio-posteriorly with minimal cortical expansion and large amount of keratin is present in cystic aspirate. Residual cyst is associated with missing teeth with history of extraction. AOTs are more common in anterior maxilla whereas central giant cell lesion are present anterior to mandibular molar.[6] Solid ameloblastoma is multilocular and uncommon in patient less than 30 years. Unicystic ameloblastoma is difficult to differentiate from dentigerous cyst. Defect in wall of cyst, unilocular cystic lesion extending into the ramus, expansion of buccal and lingual cortex favours' the diagnosis of unicystic ameloblastoma. The present case favoured the provisional diagnosis of residual cyst it was symptomatic and associated with missing first molar. Various treatment modalities for unicystic ameloblastoma have been used such as enucleation and curettage, marsupialization followed by second stage surgery and segmental or marginal resection.[9] 10-20% recurrence has been reported after enucleation and curettage.[12]

Conclusion

Unicystic ameloblastoma is a tumor with a strong tendency for recurrence, especially when the ameloblastic focus penetrates the adjacent tissue from the wall of the cyst. The diagnosis of unicystic ameloblastoma should be made by clinical, radiographic and histopathologic features. Early

diagnosis and intervention may improve the prognosis. In present case the postoperative healing was uneventful and recurrence was not noted after 6 months. It should be emphasized that every periapical pathology should be evaluated properly, both clinically and histopathlogically because despite a clinical diagnosis of residual cyst, a unicystic ameloblastoma (odontogenic neoplasm) may be present, as evident in this case.

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