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DENTIGEROUS CYST TURNING INTO UNICYSTIC-AMELOBLASTOMA: A RARE CASE REPORT

Dental Science

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ABSTRACT

Dentigerous cyst is the 2nd most common odontogenic cyst which is associated with the crown of an unerupted tooth. It is usually found in the 2nd and 3rd decades of life, but the frequency in children till now has been reported in dental literature is low. Dentigerous cysts(DC) has more potential to convert into ameloblastoma, squamous cell carcinoma, and mucoepidermoid carcinoma. Here we report a case of an 8-year-old male patient with dentigerous cyst transforming into unicystic ameloblastoma.

KEYWORDS

Dentigerous, Ameloblastoma, Maxilla, Unicystic.

INTRODUCTION:

The odontogenic cysts with the neoplastic potential include dentigerous cyst (DC), odontogenic keratocyst, calcifying odontogenic cyst, glandular odontogenic cyst, and radicular cyst. Among the odontogenic cysts, neoplastic transformation is highest in odontogenic keratocyst and DC.[1]

Ameloblastoma is a benign odontogenic neoplasm which frequently affects the mandible, and also seen in maxilla. The word ameloblastoma includes various clinico-radiological and histological variants. Apart from the most commonly encountered clinico-pathological models, there are few variants, whose biological profile is unknown or not elicited. Among these types, unicystic ameloblastoma is the least encountered variant of the ameloblastoma.[2]

Fifteen percent to twenty percent of all unicystic ameloblastomas form in the wall of dentigerous cysts. Since 1925, many had reported the development of ameloblastomas within the walls of odontogenic cysts, among which most commonly cited were dentigerous cysts.[3]

CASE REPORT

An 8-year-old male patient with his parents reported with a chief complaint of swelling in the upper front tooth region for past six months, which was initially small in size and gradually progressed to attain its present size and the swelling was not associated with pain. Systemic evaluation revealed no abnormality.

On extraoral examination, diffuse swelling of size approx 3X2.5 cms was noticed in the right middle 1\3rd of face extending anteroposteriorly from right nostril to 4cm away from the tragus of the ear causing obliteration of nasolabial fold, superior-inferiorly 2cm below infra orbital margin to 1cm below the ala-tragus line. The swelling is non-tender and is hard in consistency.

Figure 1. Extraoral facial asymmetry on right side of face Intraoral examination revealed a mixed set of dentition with retained deciduous teeth 51, 52 and non vital tooth with 51. Grade I mobility of 51, 52, 53 was also noticed. Based on these findings, a clinical provisional diagnosis of radicular cyst was given.

Figure 2. Intra oral pictures revealed diffuse swelling in the upper right buccal vestibule and palate was noticed.

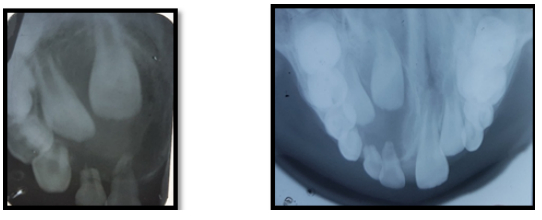

Figure 3. IOPA and cross sectional occlusal radiograph revealed well defined unilocular radiolucency attached at the CEJ of unerupted tooth(11).

A panoramic radiograph revealed a unilocular radiolucency with well-defined sclerotic border was noticed with unerupted tooth irt 11,12; extending superoinferiorly from the level of the maxillary sinus wall to the maxillary alveolus causing root resorption with 51,52. The permanent canine tooth bud was displaced.

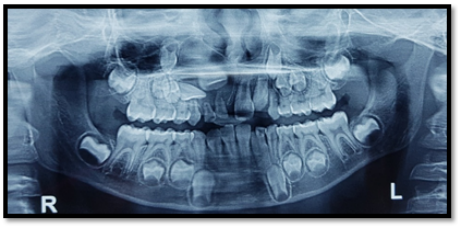

Figure 4. Orthopantomograph revealed unilocular radiolucency on right side extending from mesial aspect of 21 to mesial aspect of 14.

Computed tomography scan revealed osteolytic expansile lesion with well-defined hypodense area surrounded by hyperdense area displacing the maxillary sinus and lateral wall of nose.

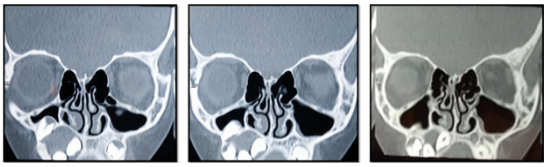

Figure 5. Bony window of CBCT coronal sections revealed well defined hypodense area on the right side of the maxilla.

Based on clinical and radiographic examination, differential diagnosis is given as dentigerous cyst, adenamatoid odontogenic tumor and ameloblastoma.

The complete lesion was surgically removed along with normal tissue margins surrounding the lesion, and the specimen was sent for histopathological examination.

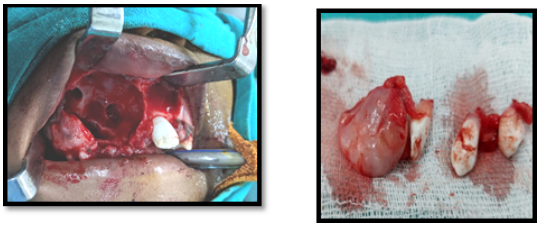

Figure 6. Enucliation of the lesion.

H&E stained sections showing cystic lumen lined by single layered cuboidal basal cell layer having hyperchromatic nucleus and superficial cells show intra and intercellular spacing resembling stellate reticulum like tissue. In few areas epithelium shows proliferation into lumen. Connective tissue is delicate to dense with haphazardly arranged collagen fibers along with intense chronic inflammatory cells, suggestive of the diagnosis of DC transforming into ameloblastoma.

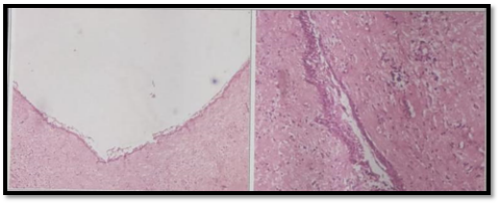

Figure 7. Histopathological report

Figure 8. Postoperative OPG

DISCUSSION

Follicular cyst is the synonym for DC. It is 2nd most common odontogenic cyst, after radicular cyst.[3] DCs were first described in France in 1778 but was not delineated until 1842 when Harris C.A published a case report on DC. It is the most common type of odontogenic cyst which is formed by the accumulation of fluid between the reduced enamel epithelium and the tooth crown and clinically associated with unerupted/impacted tooth.[4]

DCs most commonly occurs in 2nd to 3rd decade of life, according to Shear about 9% of DC occur in 1st decade of life, according to Donath, about 4% of DC appear during 1st decade of life.[4] Till date, very few cases of ameloblastoma arising from DC have been reported below first decade. In the present case report patient is within the first decade of the life which is rare case.[5]

The epithelium of odontogenic cysts may be transformed into odontogenic tumors like ameloblastoma and adenomatoid odontogenic tumors, or non odontogenic tumors. Various etiological factors have been proposed for the ameloblastomas arising from odontogenic cysts, including; extraction, trauma, infection, inflammation, unerupted tooth like nonspecific irritational factors, and nutritional deficiency, viral infections etc. Ameloblastoma is the most common odontogenic tumor, accounting for 10% of all such tumors.[5,6]

Ameloblastoma is a benign and a locally aggressive tumour which arises from the mandible or less commonly, from the maxilla. Among the various types of ameloblastomas, unicystic ameloblastoma (UCA) is a rare type, accounting for about 6% of ameloblastomas, which were first described by Robinson and Martinez, which refer to those cystic lesions that show clinical and radiological characteristics of odontogenic cysts, but on which histological examination, show typical ameloblastomatous epithelium which lines part of the cyst cavity, with or without a luminal or mural tumour proliferation.[1,5,6]

UCA usually occurs in 16–20 years of age group, with about fifty percent occur in the second decade of life. It shows slight male predilection in gender distribution with a male to female ratio of 1.6:1. However, when the tumor is not associated with an unerupted tooth, gender ratio is reversed to a male to female ratio of 1:1.8.[5]

Clinical examination reveals missing or unerupted tooth. Mostly they are asymptomatic and detected on routine radiographic examination. Facial asymmetry may be seen occasionally with hard swelling. Typically DCs has no pain or discomfort. Most common location is mandibular 3rd molars followed by maxillary 3rd molars and then maxillary anterior region.[4,6] In the present case, lesion was noticed in the maxillary anterior region which is the rarest site of UCA.

Radiologically, An important diagnostic point is that DC attaches at the CEJ of unerupted or impacted tooth. It has a potential to displace or resorb adjacent teeth and it displaces the associated tooth in apical direction, inferior alveolar canal in inferior direction in mandible, if it occurs in maxilla it causes displacement of the maxillary sinus.[6]

The clinical and radiographic features in most of the cases of UCA suggest that the lesion is an odontogenic cyst, predominantly dentigerous cyst. Few are not associated with impacted teeth which are referred as non-dentigerous variant. Non-impacted tooth-related cystic ameloblastoma mean age was 5 years, and impacted tooth-related variant is sixteen and half years. This present case report it is impacted tooth related cystic ameloblastoma of eight years old, which is a rare entity.[7,8]

Eversole et al. and Paikkatt et al. identified the predominant radiological patterns for UCA as: unilocular, scalloped macromultilocular, pericoronal, interradicular, or periapical expansile radiolucencies, in our case study peculiar unilocular appearance was noticed.[4,9]

UCA refers to those cystic lesions that show clinical, radiographic, or gross features of a jaw cyst but on histologic examination shows a typical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and/or mural tumor proliferation.[8,9]

A definitive diagnosis of UCA can only be done by histological examination of the entire lesion and cannot be predicted pre-operatively on clinical or radiographic grounds. As pre-operative incisional biopsy is not representative of the entire lesion, it may result in an incorrect classification. The epithelial lining of a UCA is not always uniformly characteristic and is often lined partly by a non-specific thin epithelium that mimics the dentigerous cystic lining.[4,9]

Histological examination reveals a 2–4 cell thickness, flat or cuboidal epithelium usually nonkeratinized, with a thin fibrous cyst wall.[5,8]

According to the World Health Organization, ameloblastomas are classified into four groups: solid/multicystic, extraosseous/peripheral,

desmoplastic, and unicystic.[9] Histopathologically, it occurs in six patterns: follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic.[10]

According to Vicker and Gorlin, the representative features of early ameloblastomatous changes include an epithelial lining parts which show transformation to cuboidal or columnar basal cells with hyperchromatic nuclei, nuclear palisading with polarization, cytoplasmic vacuolization with intercellular spacing, and subepithelial hyalinization.[11] The above findings were similar with histological features of the present case.

Histologic subgrouping by Philipsen and Reichart described as:
Subgroup 1—luminal UA;
Subgroup 1.2—luminal and intraluminal;
Subgroup 1.2.3—luminal, intraluminal and intramural;
Subgroup 1.3—luminal and intramural.
Subgroups 1 and 1.2 may be treated conservatively (careful enucleation), whereas Subgroups 1.2.3 and 1.3 should be treated aggressively. The histological typing of the current case was 1 and hence, the lesion was treated conservatively with careful enucleation.[12,13]

The recurrence rate for UAs after conservative surgical treatment (curettage or enucleation) is generally reported to be 10–20% and on average, less than 25%. This is considerably less than 50–90% recurrence rates which are noted after the curettage of conventional solid or multicystic ameloblastomas. Lau and Samman reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialisation followed by enucleation.[12,13,14]

UCA is believed to be less aggressive. As this tumor shows considerable similarities with DCs, both clinically and radiographically, the biologic behaviour of this tumor group was reviewed. Moreover, recurrence of UCA may be long delayed, and a long-term postoperative follow up is essential for proper management of these patients.[12,15,16]

CONCLUSION:

To conclude, DCs are associated with an unerupted permanent tooth is not uncommon. On rare occasions, some untreated DCs have the potential to transfer into odontogenic tumors such as ameloblastoma and malignancies such as oral squamous cell carcinoma and mucoepidermoid carcinoma. So early detection of the cyst and early treatment strategies should be started to prevent the condition or to decrease the morbidity.

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