DENTIGEROUS CYST TURNING INTO UNICYSTIC AMELOBLASTOMA: A RARE CASE REPORT

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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. 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. 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.

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 1/3 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.

On intraoral examination, 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 1. Extraoral facial asymmetry on right side of face

Intraoral examination revealed a mixed set of denition 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 irr 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.
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.

Based on clinical and radiographic examination, differential diagnosis is given as dentigerous cyst, adenomatoid 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.

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.

Ameloblastoma is a benign and a locally aggressive tumor 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. Ameloblastoma is a benign and a locally aggressive tumor 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.4,5

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 3rs molars followed by maxillary 3rs molars and then maxillary anterior region. 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.

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.6,7

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

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.

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 incisonal 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.4,5

Histological examination reveals a 2–4 cell thickness, flat or cuboidal epithelium usually nonkeratinized, with a thin fibrous cyst wall.7,11

According to the World Health Organization, ameloblastomas are classified into four groups: solid/multicystic, extrasosseous/peripheral,
desmoplastic, and unicystic. Histopathologically, it occurs in six patterns: follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic.

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, cytoplasmatic vacuolization with intercellular spacing, and subepithelial hyalinization. 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. 

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.

UCA is believed to be less aggressive. As this tumor shows considerable similarities with DCs, both clinically and radiographically, the biological 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.

**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.

**REFERENCES:**