Case Report

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Pathology Section

Hybrid Odontogenic Tumour Presenting as Unicystic Ameloblastoma with Calcifying Odontogenic Tumour: A Rare Case Report with Review of Literature

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ABSTRACT

Tumours originating from the odontogenic apparatus exhibit a high level of diversity and complexity. They exhibit a wide range of morphologies because this tissue is formed by time-dependent, closely regulated interactions between mesenchymal and epithelial elements. Notably, hybrid lesions—which consist of two or more separate regions displaying distinctive morphologic traits of different entities—are uncommon and, when they do occur, provide a diagnostic difficulty. Despite the identification of numerous hybrid lesions, their prevalence and combinations remain unknown. Ameloblastoma with Calcifying Epithelial Odontogenic Tumour (CEOT) is an extremely rare hybrid tumour, with five cases reported previously. The current paper reviews the pertinent literature and describes one more instance of a hybrid odontogenic tumour composed of ameloblastoma with CEOT. In the present case, the lesion in the mandible was provisionally diagnosed as a dentigerous cyst based on clinical and radiographic findings. But based on the histopathologic evaluation of the incisional biopsy specimen, the diagnosis of unicystic ameloblastoma with intramural proliferation was given. However, histopathology of the excised tumour revealed areas of plexiform ameloblastoma with mural proliferations along with areas of CEOT. Hence, a final diagnosis of hybrid odontogenic tumour of unicystic ameloblastoma with CEOT was confirmed. Hybrid odontogenic lesions create a diagnostic challenge. Diverse differentiation and intricate inductive connections are possible in odontogenic tumours that arise from odontogenic epithelium. Thus, it may be said that pluripotent odontogenic epithelium simultaneously causes the development of such disparate histopathological patterns inside a single tumour, resulting in the development of a hybrid tumour.

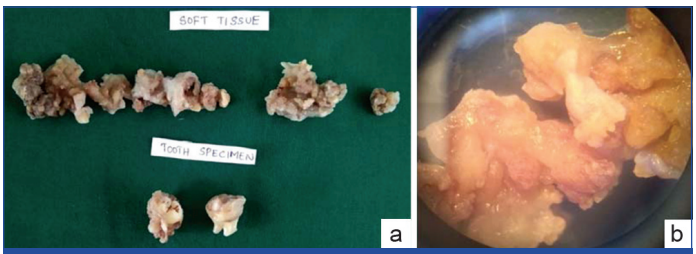
Keywords: Combined odontogenic tumour, Inductive interactions, Mixed odontogenic tumour, Pindborg tumour

CASE REPORT

A 16-year-old male patient reported with the chief complaint of pain on the right side of face for 15 days. The past medical history was insignificant and there was no history of trauma. There was a swelling of 4×3 cm size causing asymmetry of the face [Table/Fig-1a]. On palpation, the swelling extended anteroposteriorly from the angle of the mouth to the anterior border of the ramus and superoinferiorly from the subcanthal region to the lower border of the mandible. Intraoral examination revealed buccal plate expansion with perforation and missing permanent second and third molars [Table/Fig-1b]. Orthopantomogram revealed a unilocular radiolucency extending from the right first premolar to the ramus, with radiopaque flecks adjacent to the first molar. The permanent second and third molars were impacted and displaced. The second molar appeared to be pushed near the border of the mandible, and the third molar was pushed into the ramus with radiolucency around its crown [Table/Fig-1c]. Based on clinical and radiographic findings, a clinical provisional diagnosis of dentigerous cyst was given with differential diagnosis of Calcifying Epithelial Odontogenic Tumour (CEOT) (due to the presence of radiopaque flecks), ameloblastoma (due to the expansion of buccal cortical plates) and odontogenic keratocyst (because of the presence of unilocular radiolucency in the lower posterior mandible). An incisional biopsy was performed and the biopsied tissue was sent for histopathological examination. On gross examination, the tissue specimen appeared brownish-black in colour, soft to firm in consistency, and measured approximately 1.3×0.8 cm. On light microscopy, Haematoxylin and Eosin (H&E)-stained sections revealed a cystic epithelial lining surrounded by a fibrous connective tissue capsule.The lining was composed of odontogenic epithelium 3-4 cell layers thick consisting of tall columnar hyperchromatic and palisading basal cells showing reversal of polarity indicative of ameloblast-like cells. Superficial layers of loose polygonal cells were evident suggestive of stellate reticulum. Few intramural nodules/follicles were noted in the connective tissue capsule in a subepithelial location. These follicles showed peripheral columnar palisaded cells and central hypercellular areas. Along with a few dilated blood vessels and extravasated Red Blood Cells (RBC), a diffuse inflammatory cell infiltrate was observed, primarily composed of lymphocytes, plasma cells, neutrophils, and a few number of eosinophils. These results led to the diagnosis of 'unicystic ameloblastoma with intramural proliferation', and the patient was recommended to have the lesion surgically removed. However, the patient subsequently reported one year later for surgery.

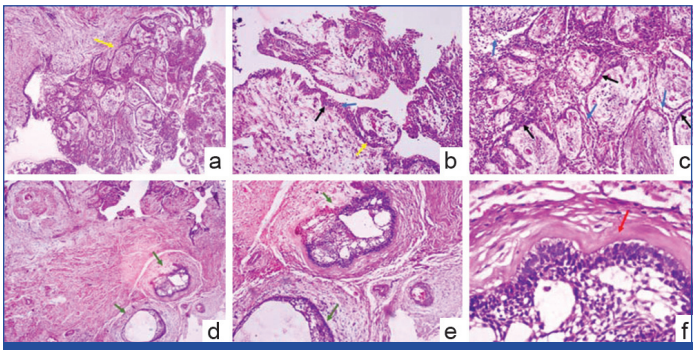
The lesion was then surgically excised (conservative enucleation and curettage), and the excised tissues were sent for histopathological evaluation. On gross examination, multiple tissue specimens were received, appearing greyish-brown in colour and soft to firm in consistency, measuring approximately 1.2×1.2 cm and 3×2 cm [Table/Fig-2a]. Under a stereomicroscope, multiple proliferations were observed on tissue specimen [Table/Fig-2b]. Histopathological analysis of tissue sections stained with H&E revealed unicystic ameloblastoma with areas of intramural proliferations. The intramural and mural proliferations of ameloblast like cells were arranged in interconnecting strands and cords with peripheral palisaded cells and central stellate reticulum cells suggestive of plexiform pattern of ameloblastoma. Areas of follicular ameloblastoma with cystic degenerations were also noted. Eosinophilic hyalinised areas suggestive of induction were evident in sub-epithelial and peri-follicular locations [Table/Fig-3].

[Table/Fig-1]: a) Extraoral swelling causing asymmetry of the face; b) Intraoral clinical photograph of the tumour in the lower posterior region; c) Unilocular radiolucency extending from lower right first premolar region till ramus of mandible. Impacted and displacked permanent right second and third molars.



[Table/Fig-2]: a) Gross specimen showing multiple soft tissue and teeth specimen; b) Stereomicroscopic picture showing soft tissue specimen consisting of numerous proliferations.

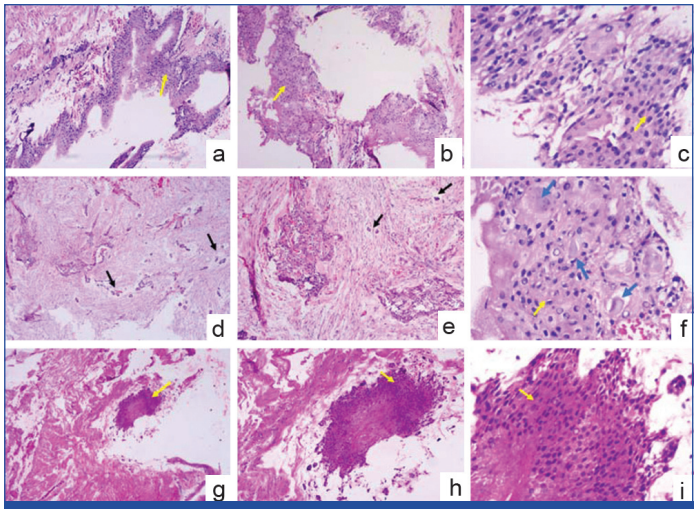
Ectomesenchyme like features with odontogenic rests were also noted in the connective tissue capsule. The connective tissue capsule exhibited predominantly myxomatous ectomesenchyme like areas along with fibrous areas. Multiple areas showed polyhedral hyperchromatic pleomorphic epithelial cells with prominent intercellular bridges with focal areas of homogeneous concentric eosinophilic hyaline masses suggestive of amyloid like material/ calcifications that are lying in the connective tissue capsule and in areas are seen lining the cystic capsule [Table/Fig-4]. A final diagnosis of hybrid odontogenic tumour—unicystic ameloblastoma with CEOT—was made. The patient was symptom free post operatively at the follow up after 9 months with no signs of recurrence.



[Table/Fig-3]: Histopathological findings: H&E staining, 40X(A), 100X(B), 100X(C). Photomicrographs depicting histopathology of unicystic ameloblastoma with intramural proliferation in the connective tissue capsule (yellow arrows). Lining consisting of tall columnar hyperchromatic and palisading basal cells indicative of ameloblast-like cells (black arrow). Superficial layers of loose polygonal cells suggestive of stellate reticulum (blue arrow). The mural proliferations of ameloblast like cells arranged in interconnecting strands and cords with peripheral palisaded cells (black arrows) and central stellate reticulum cells (blue arrows) suggestive of plexiform pattern of ameloblastoma.
Histopathological findings: H&E staining, 40X(D), 100X(E), 400X(F). Areas of follicular ameloblastoma with cystic degenerations (green arrows). F, Eosinophilic hyalinized areas suggestive of induction in peri-follicular location (red arrow).

DISCUSSION

Odontogenesis is a lengthy and intricate process, and Tumours originating from epithelial, ectomesenchymal, and/or mesenchymal components of the odontogenic apparatus are referred to as



[Table/Fig-4]: Histopathological findings: H&E staining showing photomicrographs at 40× (a, d, g), 100× (b, e, h), and 400× (c, f, i) magnifications. Polyhedral, hyperchromatic, pleomorphic epithelial cells with prominent intercellular bridges (yellow arrows) are evident, along with focal areas of homogeneous concentric eosinophilic hyaline masses suggestive of amyloid-like material/calcifications (blue arrows). Areas showing ectomesenchyme-like features with odontogenic rests in the connective tissue capsule are also observed (black arrows).

Odontogenic Tumours (OT). Thus, OTs are a wide range of lesions with different pathohistological and clinical features. These biological features include aggressive malignant Tumours, non-aggressive benign tumours, and hamartomatous proliferation [1]. There are reports in the literature of OTs that manifest as a conglomeration of established entities. Such an event may be a combined/hybrid odontogenic tumour or a collision tumour. Hybrid odontogenic tumours are defined by the coexistence of histological features of two or more odontogenic tumours in a single lesion. Unlike collision tumours, which involve independent neoplasms meeting at a boundary, hybrids arise from a common odontogenic origin and show intermingling of components [2]. Correct recognition is crucial to avoid misdiagnosis as mixed odontogenic tumours or metaplastic changes.

Literature occasionally reports tumours that co-occur, including Adenomatoid Odontogenic Tumour (AOT) with CEOT [3,4], ameloblastoma with CEOT [5], ameloblastoma with ameloblastic fibroma [6], AOT with ameloblastic fibro-odontoma [7], and ameloblastoma with glandular odontogenic cyst [8]. These cases underscore the variability in histological presentation and the importance of thorough histopathological examination in diagnosing and managing such tumours.

Ameloblastomas account for approximately 1% of all tumours and cysts of the jaws. Owing to their contradictory and paradoxical nature—being benign tumours of odontogenic epithelial origin yet exhibiting aggressive clinical behaviour with a high propensity for recurrence—they have attracted considerable attention over the years. These tumours show no gender predilection and can occur across a wide age range, with a mean age of presentation of around 33 years. Approximately 85% of cases occur in the mandible, most commonly in the molar-ramus region. Numerous histologic subtypes of ameloblastoma, including follicular, plexiform, basaloid, acanthomatous, and granular variations, have been thoroughly documented in the literature and are recognized by the World Health Organisation [9].

The rare, slowly expanding, expansile CEOT, also known as the Pindborg tumour, is made up of odontogenic epithelium devoid of odontogenic mesenchyme. Although it had previously been recorded under different pseudonyms, JJ Pindborg was the first to classify it as a distinct entity in 1955. CEOT has no gender or ethnic preference, makes up around 1% of all odontogenic tumours, and has a broad age range with a mean of 43.5 years. CEOT typically manifests as a benign tumour with minimal symptoms, but depending on the size

Author/Year	Age/Sex	Site	Clinical features	Radiographic findings	Ameloblastoma type	CEOT features	Treatment/ Outcome
Seim P et al., [5]	55/F	Right maxilla	Mild swelling, incidental finding	Radiolucency	Follicular + Plexiform + Desmoplastic	Amyloid deposits	Surgical excision, no recurrence (2 yrs)
Etit D et al., [11]	32/F	Left maxilla (posterior, sinus involvement)	Swelling	Destructive radiolucency	Follicular	CEOT nests, amyloid	Resection, no recurrence (5 yrs)
Wadhwan V et al., [12]	65/M	Right maxilla (posterior)	Swelling	Large radiopaque mass	Plexiform	CEOT sheets, amyloid	Patient declined treatment
Oruganti VR et al., [13]	40/F	Anterior mandible, crossing midline	Ulcerated swelling	Radiolucency, displacement of premolars	Unicystic granular cell	CEOT-like cells	Excision, no recurrence (3 yrs)
Ekrampoor MS et al., [14]	35/M	Anterior mandible	Swelling	Radiolucency	Follicular	CEOT cells, calcifications, vascular malformation	Recurrence after 10 yrs
Present case	16/M	Right posterior mandible	Pain, swelling, facial asymmetry	Unilocular radiolucency with flecks of radiopacity	Unicystic with mural proliferation (plexiform + follicular)	CEOT-like cells, amyloid + calcifications	Excision, follow-up ongoing

[Table/Fig-5]: Comparative analysis of reported cases: Only five cases of hybrid lesions of ameloblastoma with CEOT have previously been reported in the literature. Their clinicopathological features, in comparison with the present case, are summarised [5, 11-14].

of the lesion and its connection to nearby structures, including the maxillary sinuses, it may cause swelling and generalised pain [10].

Only five cases of hybrid ameloblastoma with CEOT have been documented to date [5,11-14]. This paper reports an additional case and reviews the existing literature [Table/Fig-5] [5,11-14].

Our case represents the sixth and is unique for being the youngest patient (16 years) reported. Previous cases occurred in adults aged 32–65 years. Both jaws are affected equally, though the maxilla was more often posteriorly involved, whereas our case involved the posterior mandible.

Clinically, swelling was the common presentation, occasionally with ulceration [13]. Radiographically, lesions were usually unilocular or multilocular radiolucencies with cortical expansion, though one case appeared radiopaque [12]. Our case showed a unilocular radiolucency with flecks of radiopacity and buccal plate perforation.

Histologically, ameloblastoma components included follicular, plexiform, desmoplastic, granular cell, and unicystic variants. CEOT areas consistently showed nests of polyhedral epithelial cells with pleomorphism, intercellular bridges, and eosinophilic amyloid-like deposits. Calcifications, rare among reported cases, were observed in only one prior report [14] and in the present case. This suggests variable differentiation within CEOT areas, possibly linked to dystrophic or cementum-like calcification [15].

Treatment across reported cases was primarily surgical excision, with no recurrences on follow-up of 2-5 years, except one case associated with vascular malformation that recurred after 10 years [14]. Our patient underwent complete excision, with follow-up ongoing.

Due to their capacity for diverse differentiation and concurrent inductive reciprocations, odontogenic tumours typically tend to develop from odontogenic epithelial remnants. One theory is that the pluripotent odontogenic epithelium may cause several histological spectrums to emerge in the same tumour at the same time [2]. Both diagnosis and therapy are extremely difficult in these circumstances. Tumour cells in hybrid tumours show immunopositivity for CK 7, CK 19, CK 8/18 and low Ki67, p63, and negative for S100 [16]. More thorough investigation is required to enhance our comprehension and treatment of these intricate and uncommon disorders, particularly at the molecular, genetic, and immunohistochemistry levels. Generally, hybrid odontogenic tumours exhibit biological behaviour similar to other solid ameloblastomas [12]. Nevertheless, the best treatment strategy remains undetermined due to the uncertain long-term behaviour of this particular hybrid neoplasm. While enucleation and excision appear to effectively treat the hybrid lesion, additional cases and extended follow-up data are necessary to verify the clinical significance and recurrence rate of these lesions.

CONCLUSION(S)

The histological characteristics of two or more known tumour types are combined in hybrid odontogenic tumours. Pluripotent odontogenic epithelial cells are thought to have the ability to produce several tissue patterns in a single lesion at the same time, yet their precise source is still unknown. CEOT and ameloblastoma could have most likely originated from the same or a similar odontogenic epithelial source, such as the dental lamina or the primitive enamel organ. It is possible for a single tumour to exhibit the histological features of both neoplasms, which is something we should anticipate. The optimal treatment strategy for this particular hybrid tumour remains undetermined due to the uncertainty surrounding its long-term behavior. Future research in molecular biology, genetics, and immunohistochemistry will broaden the scientific understanding in this field.

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