Case Report

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# UNICYSTIC AMELOBLASTOMA ASSOCIATED WITH A COMPOUND ODONTOME: AN UNUSUAL PRESENTATION

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## ABSTRACT

**Aim:** Association of odontomes with odontogenic neoplasms is reported but their manifestation in conjunction with unicystic ameloblastoma is very unusual.

**Case Report:** Herein we report a very rare case of a 28 year old male patient who was diagnosed with unicystic ameloblastoma associated with a compound odontome.

**Discussion:** Ameloblastoma is the most common true odontogenic neoplasm in the oral cavity subsequent to odontomes. Unicystic ameloblastoma is a lesser aggressive variant of ameloblastoma as compared to its solid, multicystic counterpart. Its association with odontomes has been seldom reported in English literature.

**Conclusion:** Reporting of such rare entities achieved through a comprehensive clinico pathological correlation can assist in a definitive diagnosis of similar cases.

**Key Words:** Ameloblastoma, Odontoma, Odontogenic cysts, Tooth

## INTRODUCTION

Unicystic ameloblastoma is one of the four variants of ameloblastomas, the others being intraosseous, infiltrative and peripheral type. It accounts for 5%-22%[1] of all the types of ameloblastomas but has a lower recurrence rate as compared to the other types (6.7%-35.7%)[1]. Owing its low recurrence rate, a more conservative surgical approach like enucleation or curettage or cautery is employed. Radiographically, it mimics non-neoplastic cysts which can pose diagnostic difficulties to both the surgeon and pathologists. Odontomes are most common benign hamartomas consisting of dentin, cementum or pulp like tissue found in the oral cavity[2]. Unicystic ameloblastoma occurring with an odontome is a very unusual presentation and has rarely been reported in English literature. We report a very rare case of a unicystic ameloblastoma associated with a compound odontome in a young patient.

## CASE REPORT

A 28 year old male patient reported to our institution with a chief complaint of pain in the lower right back region since two months. The swelling was insidious in onset, associated with chronic dull pain and had grown slowly to the existing dimensions. The patient did not report with any history of trauma or altered taste sensation. His past medical and dental history were non-contributory. Following this, a thorough clinical examination was carried out to determine the nature of the lesion.

Extraoral examination was normal with no visible facial asymmetry. Intraoral examination revealed a small swelling 3.5 x 2.5 x 2 cm in maximum dimensions in the parasymphseal region approaching the midline. The vestibular space in 31-35 region was obliterated. The overlying mucosa appeared normal with no ulceration and pus

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discharge. On palpation, the swelling was bony hard in consistency extending in the vestibular region from 35 to 41 region of mandible crossing the midline. The teeth associated with the swelling were non-tender, vital with no signs of mobility.

The clinical examination was followed by a complete radiographic evaluation of the lesion. Orthopantamogram (OPG) of the lesion revealed a well-defined unilocular radiolucent lesion in the mandible in relation with 35 to 43 region crossing the midline with sclerotic borders in medial and inferior aspect (Fig. 1). A few tooth like radio opacities were seen within the radiolucency in the antero-inferior region of mandible. Based on the clinico – radiographic findings, dentigerous cyst and unicystic ameloblastoma were considered in the differential list of provisional diagnoses.

Taking into consideration the clinical and radiographic findings which were suggestive of a benign cystic lesion, surgical enucleation under general anaesthesia was considered as the preferred surgical approach. The enucleated specimen was sent for histopathological evaluation. The post-operative healing was uneventful and the patient was kept under follow up.

Gross examination of the surgical specimen revealed the presence of a cystic lesion attached to the neck of a tooth like structure 3 x 1.5 x 2 cm in maximum dimensions, soft to firm in consistency, reddish brown in colour and with a smooth surface (Fig. 2). The cross section of the soft tissue specimen revealed other bits of tooth like material attached to the main tooth like structure thus establishing it to be a compound odontome (Fig. 3). The soft tissue specimen was dissected from the hard tissues and sent for routine tissue processing.

On microscopic examination the H & E stained, soft tissue section revealed a cystic lumen lined by 2-3 layers of tall columnar hyperchromatic cells with nuclei showing reverse polarity and basilar cytoplasmic vacuolisations resembling ameloblasts (Fig. 4). The superficial layer of cells were loosely cohesive and resembled stellate reticulum. Squamous metaplasia of the cystic epithelium was noted at a few places. Areas of hyalinisation in sub-epithelial region were evident in few places of the connective tissue stroma. The underlying cystic wall appeared delicate to dense fibrous with abundant areas of haemorrhage. Few odontogenic rests and dystrophic calcifications were also seen. Collections of eosinophilic material were also noted (Fig. 5). Correlating the chronic, benign nature of the cystic lesion with the unilocular radiographic appearance, a final diagnosis of unicystic ameloblastoma associated with a compound odontome was given.

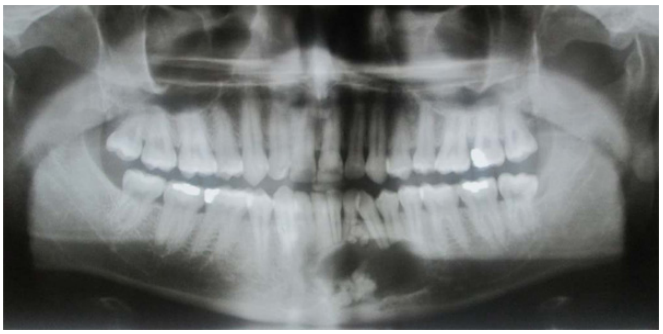

**Figure 1:** Orthopantamogram showing a well-defined unilocular radiolucency enclosing a compound odontome


**Figure 2:** Gross specimen showing cystic lesion attached to a tooth- like structure

## DISCUSSION

Ameloblastoma is the most common odontogenic tumor arising from the epithelial component of an embryonic tooth bud affecting generally molar-ramus region of the mandible[3]. Based on the clinical and prognostic aspects, ameloblastoma was characterised into[4] conventional (classic / intraosseous / solid / multicystic), unicystic and peripheral type. Unicystic ameloblastoma was first described by Robinson and Martinez in 1977. The most common site of occurrence is posterior mandible followed by the parasymphseal region. Reports from earlier literature have revealed occurrence of bone, dentin and dentinoid like material in this tumor. Ackermann in 1988

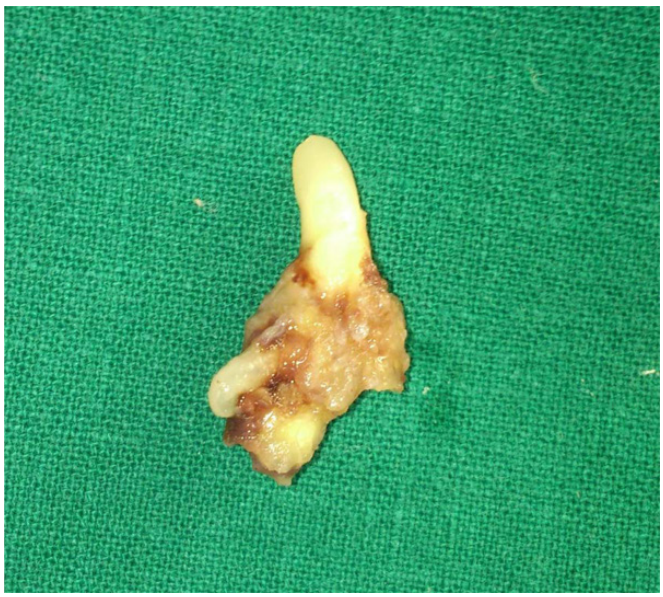

**Figure 3:** Compound odontome separated from the soft tissue specimen

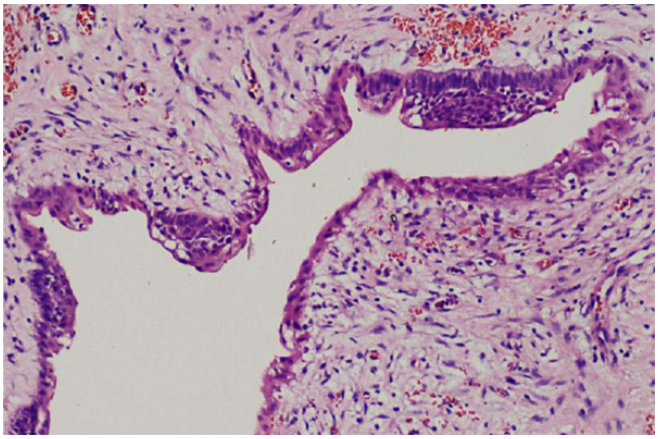

**Figure 4:** Cystic lining showing ameloblasts – like basilar cells

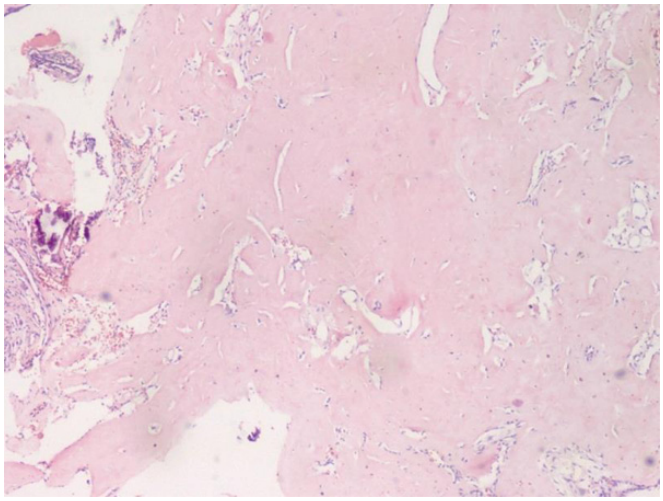

**Figure 5:** Eosinophilic material and dystrophic calcifications seen in the cystic wall

reclassified unicystic ameloblastoma into three types based on its prognostic and therapeutic implications[5, 6] which include:

1. Luminal unicystic ameloblastoma - Unilocular cyst lined by epithelium.
2. Intra luminal or plexiform type – epithelial nodules arising from cystic lining project into cystic epithelium.
3. Mural type – showed presence of invasive islands of ameloblastomatous epithelium into connective tissue wall.

Robinson and Martinez proposed unicystic ameloblastoma to be a lesser aggressive variant[7]. This feature contributes to a conservative surgical modality like enucleation of the tumor. Unicystic type was earlier thought to be a variant of solid multicystic type [4]. Our case presented with the typical findings of a 2-3 layered cystic lining with tall columnar basal cells with hyper chromatic nuclei, reverse polarity and basal cytoplasmic vacuolisation. The superficial layer had loosely cohesive stellate reticulum like cells. There were areas of squamous metaplasia. Hyalinisation in sub epithelial region in connective tissue stroma was seen in few sites. Peripherally eosinophilic material was seen. All these features were consistent with Vickers and gorlins criteria[4, 8] (1970) which led us to the diagnosis of unicystic ameloblastoma, but the presence of an associated compound odontomes was unique.

Odontoma are considered to be the most common odontogenic hamartomas found in the oral cavity. These tumors are composed of variable amounts of enamel, dentin, cementum and pulp tissue. A compound odontoma consists of agglomerates of tooth resembling material while the complex type fails to organize into the latter and consists of disordered dental hard tissues. Association of odontomes with odontogenic tumors like adenomatoid odontogenic tumour, dentigerous cyst have been reported in literature but the association of unicystic type of ameloblastoma with odontomes is very rare. As per English literature, only 2 cases of unicystic ameloblastoma, one associated with calcified hard tissues (Shivpathasundaram et al.)[9] and one associated with a single odontome by Ogunsalu et al[10] is reported. Herein we reported a very rare case of a unicystic ameloblastoma with a compound odontome.

## CONCLUSION

Unicystic ameloblastomas in association with odontomes is a very rare entity. A thorough clinical, radiographic and histopathogical examination is essential to arrive at the correct diagnosis. There is a requisite in the current scenario to report such rare cases to increase awareness among the clinicians and pathologists hence evading misperception and misdiagnosis.

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