

Neoplastic potential of odontogenic cysts

ZAMA MOOSVI, AMSAVARDANI TAYAAR S.¹, G. S. KUMAR²

Abstract

Odontogenic cysts and tumors are distinct entities and quite a common occurrence in the jaw bones. The lining of odontogenic cysts shows a potential for neoplastic transformation to non odontogenic malignancies like squamous cell carcinoma and mucoepidermoid carcinoma, and odontogenic tumors like ameloblastoma and adenoamatoid odontogenic tumor (AOT). AOT is a benign, epithelial odontogenic tumor, common site being the anterior maxilla. Its origin from a dentigerous cyst and in the mandible is rare. A case of an AOT arising from a dentigerous cyst associated with an impacted permanent mandibular left lateral incisor is reported.

Keywords: Adenoamtoid odontogenic tumor, dentigerous cyst, impacted mandibular lateral incisor, neoplastic transformation, pathogenesis

Introduction

Odontogenic cysts and tumors are distinct entities and quite a common occurrence in the jaw bones. They are individual lesions arising from the odontogenic apparatus with varying pathogenesis. The lining of odontogenic cysts shows a potential for neoplastic transformation to non odontogenic malignancies like squamous cell carcinoma and mucoepidermoid carcinoma, and odontogenic tumors like ameloblastoma and adenoamatoid odontogenic tumor (AOT).^[1] However, very few cases have been reported of tumors arising from odontogenic cysts.

In the present paper, we report a case of an AOT arising from a dentigerous cyst.

Case Report

A 13-year-old female patient visited our institute complaining of an asymptomatic swelling in the chin region, which she noticed one month back.

Department of Oral Pathology, HKE'S S Nijalingappa Institute of Dental Sciences, Gulbarga, ¹SDM College of Dental Sciences and Hospital, Dharwad, Karnataka, ²KSR Institute of Dental Sciences and Research, Tiruchengode, Tamil Nadu, India

Correspondence: Dr. Zama Moosvi, Department of Oral Pathology, c/o Dr Ali Raza Moosvi, Dean School of Earth Sciences, Central University of Karnataka, 2nd floor, Karya Soudha, Gulbarga University Campus, Gulbarga – 585 106, Karnataka, India. E-mail: zama_doc@yahoo.com

On extra oral examination, mild facial asymmetry was seen in the lower third of the face. A swelling was observed in the chin region extending superiorly to one cm below the angle of the mouth and inferiorly to two cm below the angle of the mandible with adequate mouth opening.

Intraoral examination revealed a well circumscribed swelling extending from the distal aspect of 31 to mesial aspect of 33 with normal overlying mucosa. The swelling was soft to firm in consistency and mild tender on palpation. Buccal and lingual vestibules were obliterated and Lower left lateral incisor was clinically missing. Grade II mobility of 31, 33, 41, and 42 was observed. Tongue movements were normal and there was no paresthesia of the mental region.

PNS view [Figure 1] showed a well-defined radiolucency extending from 33 to 42 regions, with a thin sclerotic border, associated with unerupted 32; 41 and 42 were displaced and 31 showed root resorption.

Provisional diagnosis of dentigerous cyst was made and incisional biopsy was done.

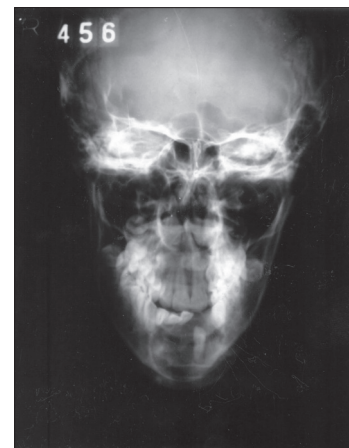


Figure 1: Paranasal sinus (PNS) X-ray view showing well-defined radiolucency in the parasymphysis region associated with unerupted 32

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Histopathology revealed a 2-3 layer thick stratified squamous non keratinized epithelium with a fibrous connective tissue wall and few inflammatory cells [Figure 2]. The findings were suggestive of a dentigerous cyst.

The cyst was enucleated under general anesthesia and 41, 42, 43, 31, and 33 were extracted. There were no operative complications and the wound healed uneventfully.

Gross examination

A well encapsulated, soft tissue mass attached to the neck of a tooth was received. The soft tissue mass was grayish in color, soft in consistency and smooth surface with regular borders. Cut section showed a cystic lumen with creamish cauliflower like proliferations from the periphery of the mass into the lumen and the crown of the tooth submerged into the lumen [Figure 3].

Histopathology

Stratified squamous to cuboidal epithelial lining of 2-3 cells thickness gradually transforming into elongated cuboidal and spindle shaped epithelial cells arranged in whorls was seen [Figures 4a-c]. Cyst wall showed fibrous connective tissue stroma arranged circumferential to the lumen with few chronic inflammatory cells.

Deeper sections were made which showed cords, whorls and few duct like patterns of round to ovoid cells in a scanty fibrillar connective tissue stroma with few areas of irregular basophilic calcifications towards the lumen [Figures 5 and 6].

The findings were suggestive of an AOT arising from the lining of a dentigerous cyst.

Discussion

Dentigerous cyst is a developmental odontogenic cyst that

encloses the crown of an unerupted tooth by expansion of its follicle, and is attached to the neck of the tooth. It is often seen in the age group of 10–30 yrs and most commonly associated with an unerupted mandibular 3rd molar or maxillary canine. It clinically presents as an unerupted tooth and / or slow growing swelling. Radiograph shows a unilocular radiolucency associated with crown of an unerupted tooth, with well-defined sclerotic margins. Histological examination shows a 2–4 cells thick, flat or cuboidal epithelium (usually non keratinized), and with a thin fibrous cyst wall.^[2]

Few cases reported in literature show the neoplastic potential of the epithelium of dentigerous cysts to ameloblastoma, AOT, and mucoepidermoid carcinoma.

AOT, first described by Stafne in 1948,^[3] is a benign, epithelial odontogenic tumor that accounts for 3–7% of all odontogenic tumours. It is usually seen in the age group of 13–30 yrs, common in females, and often located in the anterior portion of the jaws, especially the maxilla. Clinically presents as an asymptomatic swelling usually associated with an unerupted tooth (most commonly the maxillary canine). Radiograph presents it as a well defined radiolucency commonly associated with an unerupted tooth and sometimes contains faint radio opaque foci.^[4]

Broadly AOT are of two types; central and peripheral. Further, central type shows a follicular variant associated with an unerupted tooth and an extra follicular variant, which is usually placed between the roots of adjacent teeth and is not associated with an unerupted tooth. Sometimes the tumor may cause root resorption and displacement of adjacent teeth.^[5]

On histological examination, the tumor consists of whorls or nodules of spindle shaped or cuboidal epithelial cells. The cells form duct-like structures or spherical microcysts that are lined by cuboidal to columnar cells, which resemble

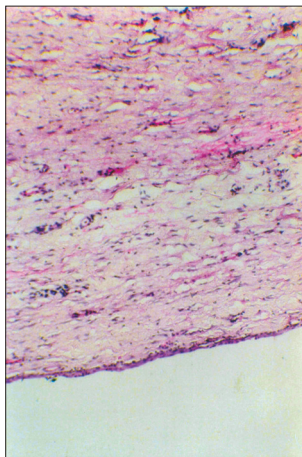


Figure 2: Photomicrograph showing thin stratified squamous non keratinized epithelial lining of dentigerous cyst with a fibrous connective tissue wall and few inflammatory cells (H and E, ×100)

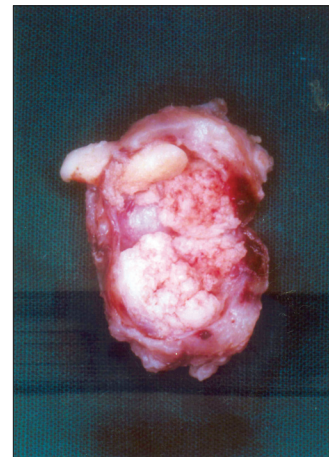


Figure 3: Cut section of the gross specimen, showing proliferations from the periphery of the mass into the lumen and mass attached to the neck of the tooth

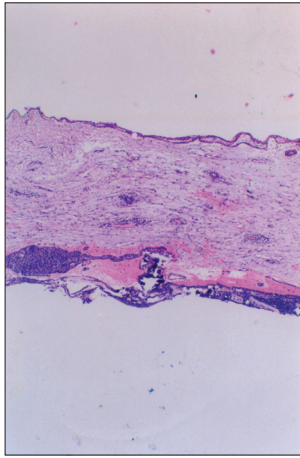


Figure 4a: Scanner view showing cyst lining gradually transforming to whorls of elongated cuboidal cells and connective tissue stroma arranged circumferential to the lumen with few chronic inflammatory cells. (H and E, x40)

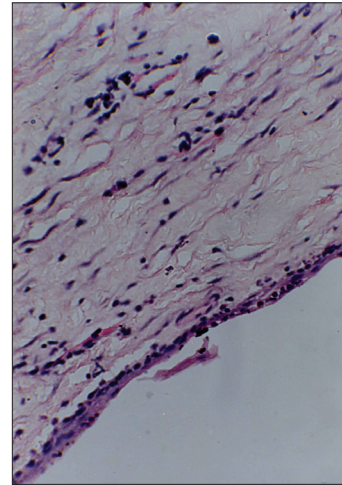


Figure 4b: Photomicrograph showing lining of ntigerous cyst and loose connective tissue wall with few inflammatory cells. (H and E, x250)

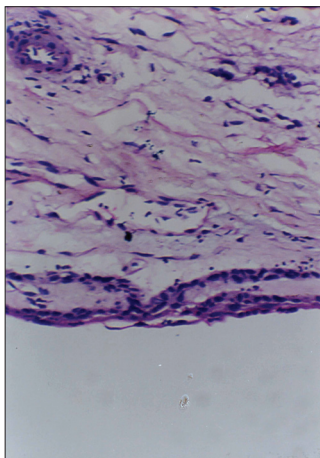


Figure 4c: Photomicrograph showing cyst lining gradually transforming to elongated cuboidal cells of AOT and connective tissue wall with few inflammatory cells. (H and E, x250)

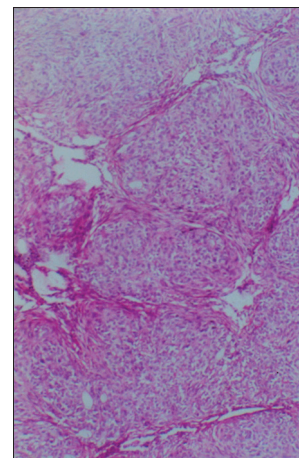


Figure 5: Photomicrograph showing cells arranged in whorls and duct like patterns with calcification. (H and E, x250)

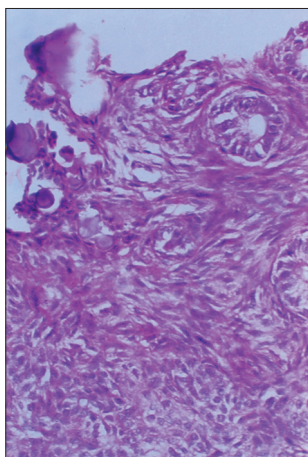


Figure 6: Photomicrograph showing cords, whorls and few duct like patterns of round to ovoid cells in a scanty fibrillar connective tissue stroma. (H and E, x100)

ameloblasts and the lumen of the microcysts contains homogenous eosinophilic material. Basophilic calcifications resembling cementum or occasionally dentinoid may be seen among the sheets of epithelial cells. Connective tissue stroma is loose with scanty cellular elements.^[6]

The histogenesis of AOT is controversial with various concepts being proposed as to its origin from fully formed enamel organ, dental lamina and/or its remnants^[11] and from odontogenic cysts.^[7,8] The tumor is managed by enucleation and recurrence is rare.^[6]

Although these are the typical features of an AOT, unusual variations in clinical and histological features have been reported, which include AOT occurring in the posterior region of the jaws,^[9] AOT occurring in combination with calcifying epithelial odontogenic tumor,^[10] AOT showing melanin pigmentation,^[11] and AOT arising from odontogenic cysts.^[7,8]

The present case shows typical features of an AOT occurring in a 13-year-old female patient, in the anterior region of the jaws and associated with an unerupted tooth. Besides the lining of a dentigerous cyst, histopathology of this case showed whorls and duct like patterns of round to ovoid epithelial cells in a scanty connective tissue stroma.

The rarity of this case lies in its histogenesis, wherein the tumor seemed to be arising from the lining of a dentigerous cyst. Further, the tumor was associated with an unerupted mandibular lateral incisor and the frequency of impaction of this tooth is rare.

Conclusions

Our present case of an AOT arising from a dentigerous cyst is a rare entity that unfolds the histogenesis of AOT. Further, the tumor occurring in the anterior portion of the mandible, associated with mandibular lateral incisor is not common.

Hence, this case highlights the neoplastic potential of odontogenic cysts and alerts us for a meticulous histopathological examination of the biopsied specimens, which can unfold the mystery behind origin of tumors.

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