

CASE REPORT

Unusual Dentigerous Cyst Undergoing Ameloblastomatous Proliferation: A Rare Case Report

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ABSTRACT

Dentigerous cyst is a commonest developmental Odontogenic cyst that grows by fluid collection over the top of an impacted crown and reduced enamel epithelium. Moreover, it is usually associated with impacted tooth and surrounding the crown of the tooth. But, this case report highlights a well-defined radiolucent lesion in the body of the mandible which is not associated to an impacted wisdom tooth or any other tooth in radiographical representation. Furthermore, histological examination demonstrates reduced enamel like epithelium and it was conformed as dentigerous cyst.

The histological feature also revealed ameloblastomatous transformation along with cholesterol clefts and calcification in the lumen which rarely occurs with the dentigerous cyst making it unique. Therefore, a conformed diagnosis is necessary that can be done by microscopical examination to take appropriate therapeutic decision.

Keywords : Dentigerous cyst, Hertwig's sheath, Vicker and Gorlin criteria, Wisdom tooth.

INTRODUCTION

Dentigerous cysts are the most prevalent developmental odontogenic cysts after inflammatory cysts such as radicular cyst which accounts for approximately 25% of all cases.

The dentigerous cysts are seldom identified in young individuals since they frequently manifest in individuals between 20 and 40 years of age, with peak incidences in teenagers (Bhushan NS et al. 2014).

Mandibular third molars are the most commonly teeth related to these type of cysts succeeded by maxillary canines, mandibular premolars, maxillary third molars and supernumerary teeth. Dentigerous cysts most commonly manifest as painless intra-oral alveolar swelling. These cysts are discovered unexpectedly on routine radiographic examination since dentigerous cysts are asymptomatic unless after an infection (Arakeri G et al. 2015). Only those teeth with a radiographic finding of peri-coronal space of less than 2.5 mm were considered. Two Oral Pathologists reviewed the slides for any changes suggestive of cystic pathology. It has greater tendency to induce root resorption of adjacent teeth (21.6%) than other jaw bone cyst such as OKC (Aher V et al. 2013).

Furthermore, Stanley et al. recommended that if the peri-coronal space around the crown of affected tooth is larger than 4mm in radiographic finding then it is regarded as small dentigerous cyst which separates from dental follicles (Saha SS et al. 2020). Some literature has emphasized that bilateral dentigerous cysts most commonly connected with certain syndromes such as mucopolysaccharidosis(type VI) and cleidocranial dysplasia whereas multiple dentigerous cysts is in Maroteaux–Lamy Syndrome (Shah A et al. 2017).

Histologically, it consists of a fibrous connective tissue wall, a lumen which is lined by nonkeratinized, stratified, squamous epithelium. The cancerous alteration is supreme in odontogenic keratocyst along with radicular cyst and dentigerous cyst. About Fifteen percent to twenty percent of entire unicystic ameloblastomas are arising from the fibrous connective tissue wall of dentigerous cysts (Rao N M et al. 2014).

Unicystic ameloblastoma were first described by Robinson and Martinez is one of the most common variants of ameloblastoma, which is a luminal lesion with clinical and radiological features of odontogenic cysts (Saluja K et al. 2010). The most common microscopical feature reveals ameloblastomatous epithelium transformation which encircles a portion of the cystic lumen with or without a luminal or mural neoplastic proliferation (Jaafari A et al. 2015).

In initial stage ameloblastic like changes comprised of cuboidal or columnar basal cell transformation with hyperchromatic nuclei, cytoplasmic vacuolization with intercellular spacing, nuclear palisading with polarisation, and subepithelial hyalinization which is according to Vicker and Gorlin criteria. The first case of ameloblastoma arising from a dentigerous cyst was reported 1933 by Cahn (Saluja K et al. 2010). Recent research has discovered that tumor associated markers such as p63, CD10, and osteopontin appear to be up-regulated in dentigerous cysts that are transforming into ameloblastoma (Gupta S et al. 2011).

So, this article is highlighting a unique case of an unicystic ameloblastoma which is emerging from the epithelial lining of dentigerous cyst. Interestingly, it is considered more especial as it is not associated with crown of the impacted tooth.

Case Report

Chief Complain

A male patient of 67year old was referred to the Department of Oral and Maxillofacial Pathology and Oral Microbiology with mild painless swelling in the lower back region for 1 year. Past dental history revealed extraction of his lower teeth 4 years back. (**Figure 1**)

Intraoral examination revealed a diffuse solitary swelling present on left side of the alveolar ridge extending from edentulous 35-38 region along with buccal and lingual cortical plate expansion. The size of the swelling was of approximately 2x1cm with smooth surface & the overlying mucosa was normal.(**Figure 2**) On palpation, inspection findings were conformed which was firm to bony hard in consistency, non-tender and non-pulsatile at the time of examination.



Figure 1: Photograph showing extraoral examination.



Figure 2: Photograph showing intraoral examination.

Radiographical Feature

Panoramic radiograph exposed a rounded, unilateral, well defined, radiolucent area in the body of the mandible which was not associated with impacted tooth. (**Figure 3**)

In addition to conventional radiographical techniques, cone beam computed tomography was recommended to determine the extent of the lesion. It provides accurate information about the size which was 2.4x1.1x1.8cm in the greatest antero-posterior, transverse and superior-inferior dimensions respectively.(**Figure 4**) Also, the margins were distinct, thin, corticated and inferior displacement of the left inferior alveolar canal. Minimal expansion was seen of the adjoining buccal and lingual mandibular cortex.(**Figure 5**)



Figure 3: Orthopantomograph revealing well defined radiolucency in the left body of the



Figure 4: CBCT images

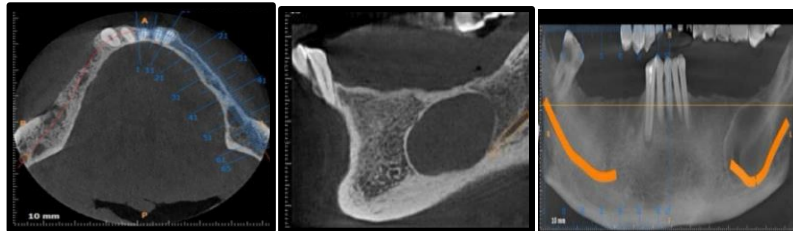


Figure 5: CBCT images (3D reconstruction) demonstrating osteolytic destruction of the mandible.

Provisionally Diagnosis

On the basis of clinical and radiographical features, provisional diagnosis for the lesion included residual cyst, OKC and radicular cyst.

Microscopical Feature

Histological examination revealed a cystic lumen which is lined by 2-3 layers of nonkeratinized squamous epithelium consisting of flattened cells surrounded by a fibrous connective tissue capsule. (Figure 6 & 7) In focal areas hyperplastic squamous epithelial lining was seen. Characteristically, epithelium showed transformation into odontogenic epithelium with reversal polarity of hyperchromatic nuclei of basal cells, loosely arranged stellate reticulum like cells and subepithelial hyalinization. Moreover, the capsule wall is fibrous with chronic inflammatory cell infiltration at some areas. (Figure 8)

Foci of haemorrhage, calcification, cholesterol clefts and few multinucleated giant cells were also interspersed within the lumen. (Figure 9) Correlating with above features an accurate diagnosis confirmation of the dentigerous cyst with ameloblastomatous transformation was conformed.

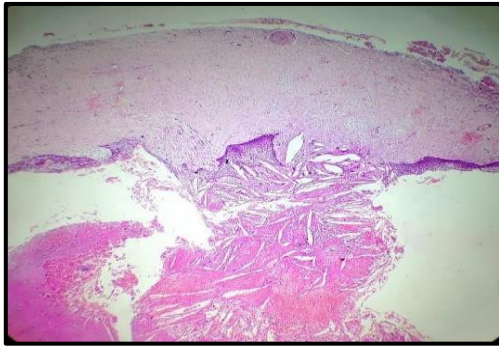


Figure 6: Histopathological image demonstrating fibrous capsule, epithelium and lumen. (H&E stain, 4x magnification)

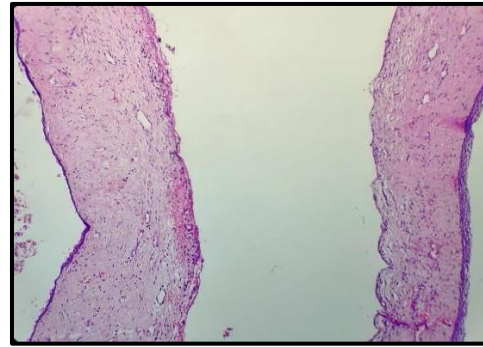


Figure 7: Histopathological image demonstrating epithelial lining. (H&E stain, 10x magnification)

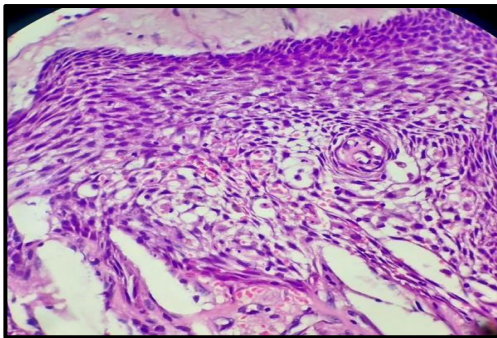


Figure 8: Histopathological image demonstrating ameloblastomatous changes in cyst with stellate reticulum like cells dentigerous. (H&E stain, 40x magnification)

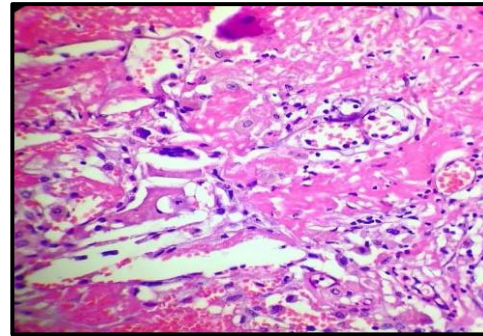


Figure 9: Histopathological image demonstrating giant cell in the epithelium. (H&E stain, 40x magnification)

DISCUSSION

It is mostly identified on the basis of its radiographical appearance associated with the tooth and retained deciduous tooth or absence of permanent teeth. But, it must be diagnosed along with microscopic examination of the specimen which is crucial in giving accurate diagnosis. Our case report was clinically diagnosed as a residual cyst due to its typical radiographical presentation as it was not associated with crown of impacted tooth which makes it very unique. Furthermore, the epithelium dentigerous of the cyst may transformed into variety of lesions, notably Ameloblastoma, Adenomatoid odontogenic tumor, Oral Squamous Cell Carcinoma, Mucoepidermoid carcinoma etc. Approximately 15% to 20% of all unicystic ameloblastomas originate from the wall of dentigerous cysts(Stanley HR et al. 1988).

Ameloblastoma is the aggressive form of benign tumors tendency for recurrences. It conceivably derived from epithelium of odontogenic cysts, particularly the dentigerous cyst and odontomas, cell rests of the enamel organ, either remnants of dental lamina or Hertwig's sheath, disturbances of the developing enamel organ, basal cells of the surface epithelium and heterotopic epithelium in other parts of the body, especially the pituitary gland (Shah A et al. 2017).

Few literature has established till now on cyst-tumor transformation. This article also demonstrated dentigerous cyst histologically transforming into unicystic ameloblastoma. Treatment include enucleation and decompression followed by enucleation if the cyst is large.

CONCLUSION

In view of ameloblastoma like epithelial transformation which is one of the most common changes potential of dentigerous cysts. Thus, it is crucial to recognize true ameloblastoma like epithelium changes of odontogenic cysts which is unrecognized in most of the cases. Sometimes, when odontogenic cysts are inflamed or no other histological diagnostic trademark of unicystic ameloblastomas are evident. Other diagnostic criteria for unicystic ameloblastomas, as represented by Vickers and Gorlin should be recommended to plan appropriate treatment.

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Conflicts of Interest

There are no conflicts of interest.

REFERENCES

1. Aher V, Fareedi B, Mukram A, Chander, Prasant C, Rajkumar G. Dentigerous cysts in four quadrants: A rare and first reported case. *J Surg Techniq Case Report*. 2013; 5(1):21–25.
2. Arakeri G, Rai KK, Shivakumar HR, Khaji SI. A massive dentigerous cyst of the mandible in a young patient: a case report. *Plast Aesthet Res*. 2015; 2:294-298.
3. Bhushan NS, Rao NM, Navatha M, Kumar BK. Ameloblastoma arising from a dentigerous cyst-a case report. *J Clin Diag Res*. 2014; 8(5):23-25.

4. Gupta S et al. Unicystic Ameloblastoma of the Maxilla in a 19 Year Old Patient- A Rare Case Report. *Ind J Stomatol.* 2011; 2(4):282-84.
5. Jaafari A, Zohreh A et al. P63 and Ki-67 Expression in Dentigerous Cyst and Ameloblastomas. *J dent .* 2015; 16(4):323-328.
6. Rao N M et al. Ameloblastoma Arising from A Dentigerous Cyst- A Case Report. *J Clin Diagn Research.* 2014; 125(2): 82-85.
7. Saha SS, Gandhoke HK, Mahato B, Deb T. Dentigerous cyst with ameloblastomatous proliferation as well as calcifications: An unusual presentation. *J Ind Acad Oral Med Radiol.* 2020; 32:297-299.
8. Saluja K et al. Multiple dentigerous cysts in anonsyndromic minor patient: Report of an unusual case. *Nation J Maxillofac Surg.* 2010;2(1):68-72.
9. Shah A, Hakim T et al. Multiple Dentigerous Cysts as a Rare Presentation of Maroteaux-Lamy Syndrome. *Annal maxillofac surg.* 2017; 7(2):282–286.
10. Stanley HR, Alattar M, Collett WK, Stringfellow HR Tr, Spiegel EH. Pathological sequelae of “neglected” impacted third molars. *J Oral Pathol Med.* 1988; 17:113- 17.