

Case Report

Dentigerous Cyst With Mucous Prosoplasia -Unusual Case Report And A Brief Review

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ABSTRACT:

Dentigerous cysts, also called follicular cysts, are developmental odontogenic cysts that occur in association with an unerupted or impacted tooth. Fluid accumulation between the enamel epithelium and enamel causes the dental follicle to separate from the crown, giving rise to such cysts. These cysts do not show any symptoms and are diagnosed during routine radiological examination. Mucous cell prosoplasia is a rare type of entity commonly seen in oral pathologies such as odontogenic cysts and might lead to diagnostic difficulties. Here we present an unusual case of dentigerous cyst with mucous prosoplasia.

Keywords: Odontogenic cyst, dentigerous cyst, mucous prosoplasia, follicular cyst

Received: 15 March, 2023

Accepted: 19 April, 2023

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This article may be cited as: Mohtesham I, Jayarajan D, Abdulla R, Adyanthya S, Moidin S. Dentigerous Cyst With Mucous Prosoplasia -Unusual Case Report And A Brief Review. J Adv Med Dent Scie Res 2023;11(5):39-42.

INTRODUCTION

The odontogenic tissue surrounding an impacted tooth is prone to develop into an odontogenic cyst or tumor. Dentigerous cyst (DC) considered the most common developmental cysts among the odontogenic cysts. The DC is defined as, “developmental” odontogenic cyst associated with the crown of an impacted tooth mostly a permanent tooth. They are invariably seen in relation with embedded, unerupted, or impacted permanent tooth crown.^[1,2] They are frequently found in association with mandibular third molars. They rarely present with any alarming symptoms and are usually discovered accidentally or incidentally on radiographs generally taken for investigation for delayed tooth.³ They may become symptomatic only when its infected and may change in size causing displacement of teeth and bone expansion⁴ Radiographically, DC exhibits a as well-defined, radiolucent lesions with sclerotic borders associated with an unerupted tooth and surrounded by a cystic space exceeding 5 mm.² Moreover a large cyst shows a multilocularity posing diagnostic dilemmas to the clinicians. Histopathologically, they manifest as a cystic cavity which is usually lined by 2-3 layered, thin nonkeratinizing epithelium that is devoid of rete

pegs, with a fibrous wall devoid of any inflammatory components. It is observed that the dental cyst arises from proliferation of the epithelial rests of Malassez in a focus of inflammation stimulated by pulpal necrosis of the associated tooth. It enlarges by unicentric expansion from the hydrostatic pressure of its contents. The DC arises from pooling of inflammatory exudate, which is derived from the obstructed follicular veins of an unerupted tooth and accumulates between the reduced enamel epithelium and the crown of the tooth. It enlarges by unicentric expansion from the hydrostatic pressure of its contents.⁴ Enucleation and extraction of the associated tooth is the preferred treatment option.³

Apparently rare but secondary complex odontome (CO),^{11,12} ameloblastoma (AB), mucoepidermoid carcinoma (MEC) and squamous cell carcinoma (SCC) have been documented in various case reports and series of cases.⁵

Cellular differentiation is an important aspect in the development of organism in which a cell changes from one type to the other. The differentiated cells are more specialized.⁶ Differentiation occurs throughout the life and has many physiological events that lead to formation of complex system. Numerous pathologies

also exhibit differentiation in the form of metaplasia, anaplasia and prosoplasia. One of the most enigmatic types of cellular differentiation in pathology is prosoplasia.⁷

Here, we report a case of the dentigerous cyst with mucous prosoplasia that presented as an incidental finding.

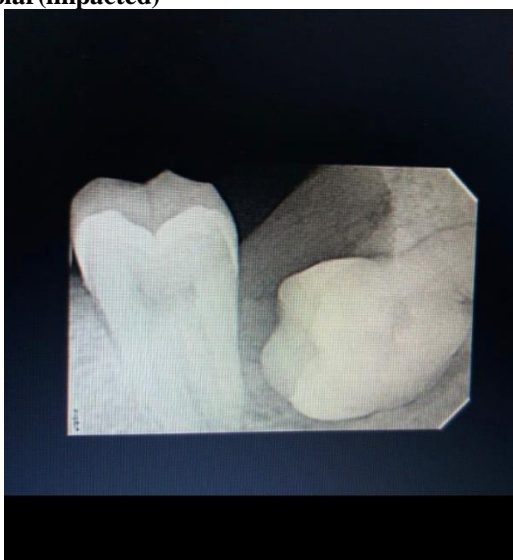
CASE REPORT

A 44-year-old male patient reported to the department for general dental check-up. Apart from several missing teeth including 38, the clinical examination revealed no obvious relevant extraoral or intraoral findings. Patient had no known medical history. Routine radiographic examination revealed an unilocular radiolucency that was well-circumscribed and involved the crown of impacted 38(Figure 1).

Table 1: Demographics and incidence of dentigerous cysts according to a retrospective analysis of 2082 cases by L. L. Zhang et al.

| | |
|---|--|
| Frequency of occurrence amongst jaw cysts | 30% of all jaw cysts |
| Common age of incidence | 2 nd – 3 rd decade |
| Predominantly affected ethnicity | Caucasian |
| Predominant gender | Males (1.6:1) |
| Commonly associated tooth | Mand. 3 rd molar > maxillary third molar > maxillary canine |

Figure 1: radiographic image of 2nd molar and 3rd Molar(impacted)



A preoperative provisional diagnosis was given as dentigerous cyst. The cyst enucleation with the extraction of the impacted wisdom teeth was done under local anesthesia. The postoperative follow-up was uneventful. The patient was advised to follow-up every 6 months.

The enucleated specimens were sent for histopathological diagnosis. Macroscopically, the soft-tissue specimens were firm inconsistency measuring

1.5x1.9 cm² and brownish-white in colour. The specimen was sent for routine H and E staining procedure. Microscopically the section showed cystic cavity lined by non-keratinised epithelial lining of 2-3 layers thickness exhibiting mucous cells suggestive of mucous prosoplasia. The connective tissue stroma was composed of parallelly arranged dense collagen fibres interspersed with chronic inflammatory cells such as lymphocytes and plasma cells. With the above mentioned features, a diagnosis of the dentigerous cyst with mucous prosoplasia was given (Figure 2 and figure 3).

Figure 2: histopathology image under 10X

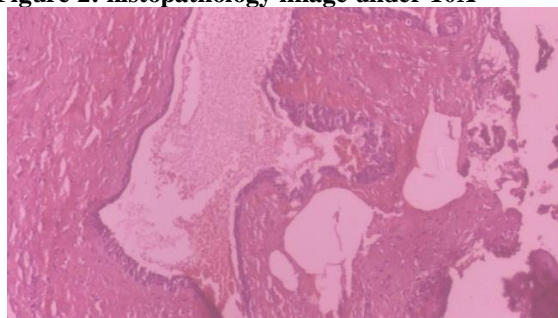
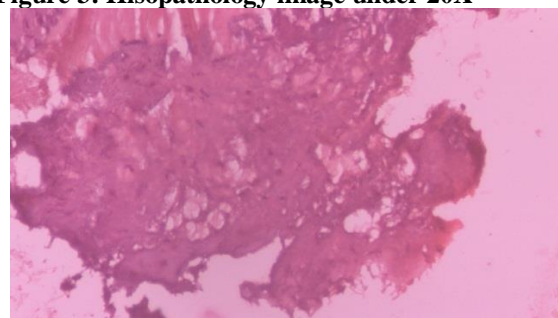


Figure 3: Hisopathology image under 20X



Based on the radiographic and microscopic features, the final diagnosis was established as dentigerous cyst with mucous prosoplasia.

DISCUSSION

Dentigerous cyst is caused because of the inflammatory exudate pooling between reduced enamel epithelium and crown of the tooth or between the layers of the enamel epithelium. The exudate results from the obstructed follicular veins of an unerupted tooth. In comparison to inflammatory cysts there is no epithelial proliferation in its development. The cyst wall is derived from the dental follicle, consisting of scattered odontogenic epithelial rests which occasionally exhibit dystrophic calcification. Mucus-producing cells, as well as ciliated cells, may be observed in the lining, which marks the multipotentiality of the cells of the dental lamina. Cyst enlarges by unicentric expansion from the hydrostatic pressure of its contents. Rarely, secondary development of neoplastic lesions such as: Adenomatoid Odontogenic Tumour, Complex Odontoma, Ameloblastoma, Mucoepidermoid Carcinoma, and Squamous Cell Carcinoma have been documented. Although the precise mechanism of malignant transformation in the lining epithelium

remains unknown, long term chronic inflammation may stimulate this transformation.

Radiographically DC often exhibits a well demarcated unilocular radiolucent lesion attached to the unerupted tooth (cervical line) with well-defined margins and sclerotic borders. A dentigerous cyst and a normal dental follicle are differentiated only based on size seen on the radiograph (a cystic space exceeding 5 mm dentigerous cyst to be suspected). Nevertheless, multilocular radiolucency may be seen in large cysts and ill-defined margins are seen in infected cysts giving rise to diagnostic dilemmas. In the present case, the typical well-circumscribed unilocular radiolucency of the dentigerous cyst, was present in relation to 38, that aided us in the incidental clinical diagnosis, even in the absence of clinical symptoms, or signs.

Prosoplasia is the word derived from two ancient Greek words, 'prósopon' meaning 'face' and 'plasis' meaning 'formation'. It is the differentiation of cells either to a higher or intricate function or to a complex level of organization. Thus, prosoplasia is basically known as development of a new complicated cell function. It is contemplated as a forward differentiation in contrast to anaplasia, which is considered as a retrograde differentiation.⁷ The presence of mucous cells in the cystic epithelium has been the subject of several theories, a few being:

- 1. Lining of the antral cavity:** Shear M proposed in 1960 that the appearance of mucous secreting cells in the lining epithelium of OCs can be attributed to the transplantation of these cells from an adjacent epithelium that ordinarily contains mucous cells, such as the nasal and antral cavity epithelial lining.¹⁰
- 2. Pluripotential embryological cells:** Mucous cells may form in the lining epithelium of OCs as a result of embryological pluripotent cells in the remaining cells from which cysts are supposed to arise, according to Shear M in 1960.¹⁰ Browne RM discovered a difference in the occurrence of mucous cells in distinct types of OCs during a histologic analysis of the walls of 638 OCs.⁴ The pluripotent embryological cell hypothesis for the formation of mucous cells in OCs is contradicted by this. If mucous cells in OCs are derived from pluripotent embryological remnant cells, their frequency of occurrence in the cystic lining of different OCs should be similar.
- 3. Theory of transdifferentiation:** Hodson JJ postulated in 1956 that the existence of mucous secreting cells in the cystic lining epithelium could be the result of prosoplastic alteration of normal lining epithelial squamous cells into mucous secreting cells in response to changes in their environment.¹¹ Toller PA's biochemical examination of the cystic contents of distinct cysts revealed that the soluble protein contents of RCs and DCs were identical, while that of odontogenic keratocysts were different.¹² As a result,

environmental inconsistencies could be to blame for the change in the occurrence of mucous cells in these cysts. In addition, the keratin layer that lines the cystic epithelium of odontogenic keratocysts may explain the low incidence of mucous prosoplasia by protecting epithelial cells from such inductive environmental stimuli. Other than the above mentioned theories, some authors also found a remarkable association between mucous and vacuolated cells.

Mucous cell prosoplasia, which is the transition of a simple squamous epithelial cell into a mucous secreting cell, is a well-known example of prosoplastic switch. Histopathologically, this condition appears as polygonal to cuboidal cells with basophilic to amphophilic cytoplasm, indicating the presence of mucin in the cell. In the epithelial linings, they are frequently found in clusters or alone.^[9] Mucous cell prosoplasia is seen in various OCs such as radicular cysts, residual cyst, DC, and odontogenic keratocyst, and in odontogenic tumors such as ameloblastomas. In 361 odontogenic cysts examined, mucous cells were present in 18.1% of radicular cysts, 23.8% of DCs and 26.9% of primordial cysts. Mucous cell prosoplasia was observed in 11.9% of dental follicle specimens in a study conducted by Cabber F et al. Having said, it is intriguing and fascinating to have the presence of prosoplastic and mucosal changes in OCs such as RCs and DCs which are not consistent with other such changes in other parts of body.

CONCLUSION

Dentigerous cyst is a common developmental odontogenic cyst. Common site of its occurrence is mandibular posterior- third molar region, and commonly associated with impacted third molar. Prosoplasia is an unusual occurrence in dentigerous cyst. Usual complication associated with dentigerous cyst: ameloblastoma, epidermoid carcinoma, mucoepidermoid carcinoma and adenomatoid odontogenic carcinoma.

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