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Infected dentigerous cyst on a totally impacted mesiodens: A rarest combination of two ectopic pathological entities

Ferdian Rizky Hutomo

Resident of Oral and Maxillofacial Surgery, Faculty of Dental Medicine, Universitas Airlangga, Surabaya, Indonesia

Andra Rizqiawan

Department of Oral and Maxillofacial Surgery, Faculty of Dental Medicine, Universitas Airlangga, Surabaya, Indonesia

*Corresponding author email: andra-r@fkg.unair.ac.id

Michelle Margaretha

Dentistry Program, Faculty of Dental Medicine, Universitas Airlangga, Surabaya, Indonesia

Dewi Cahya

Dentistry Program, Faculty of Dental Medicine, Universitas Airlangga, Surabaya, Indonesia

Abstract---Dentigerous cyst is a developmental odontogenic cyst which originates through alterations of reduced enamel epithelium in an unerupted tooth or a developing tooth bud. These cysts rarely associate with supernumerary teeth. Supernumerary teeth accounts for 5% of all dentigerous cysts. The most common supernumerary teeth are mesiodens. The case study aimed to present a rare case of infected dentigerous cyst on a totally impacted mesiodens. This case report describes a rare case of infected dentigerous cyst associated with mesiodens in a 33 year old female who presented with a slow growing swelling in the palatal. Dentigerous cyst was treated with cyst enucleation with extraction of the mesiodens. Mesiodens should be diagnosed at the earliest possible time and its management should be conducted to prevent development of dentigerous cyst and damage to adjacent teeth and vital structures.

Keywords---dentigerous cyst, health outcomes, mesiodens.

Introduction

Dentigerous cyst is a pathological space lined by epithelium or fibrous tissue follicle envelope by layered squamous epithelium surrounding the crown of an unerupted tooth (Mishra et al., 2017). This condition could occur on impacted teeth, unerupted permanent teeth, supernumerary teeth, odontomas, and primary dentitions. Five percent of dentigerous cysts are related to supernumerary teeth, such as mesiodens (Hassnain et al., 2017). Clinical examination revealed missing teeth and hard swelling in several cases, occasionally leading to facial asymmetry as well as pathological fractures. Dentigerous cysts are usually found upon routine radiographic examinations, accounting for their asymptomatic nature. Radiographically, the cyst appears as a unilocular radiolucent image, with a clear sclerotic border surrounding the crown of an unerupted tooth; however, the border of an infected cyst is diffuse (Gendviliene et al., 2017).

Mesiodens is a supernumerary tooth located at the midline between the maxillary central incisors (Reddy et al., 2018). The etiology of mesiodens is not clearly defined. The prevalence of mesiodens ranging from 0.15 to 1.9% among the population, and it has a higher predilection in males (2:1.4). Mesiodens could appear either as erupted or impacted teeth or are rarely associated with dentigerous cysts. Mesiodens tends to grow labially and eventually grows in approximation to the nasal base (Hassnain et al., 2017). The modality of treatment varies, from enucleation to marsupialization, including extracting the unerupted tooth (Reyes et al., 2018).

Case Description

A 33-years-old female presented to Hospital of Universitas Airlangga with a chief complaint of a swelling in her palate. The patient has observed the swelling for six months prior. The swelling gradually grew larger and expanded to the anterior gum and lips, with minimal pain on the swelling around the gum near the lips, the swelling has never ruptured nor shrunk. The adjacent tooth started to drift since the swelling appeared, with no history of dental pain nor fever before the swelling appeared. Upon physical examination, the patient was in good general health. The clinical examination revealed a silting of the philtrum (Figure 1) and a moderate swelling involving the vestibular region of 13 to 22 teeth and the palate (Figure 1b). The palpation of the lesion in the vestibular region shows a cystic consistency, meanwhile a hard consistency in the palatal region. The mobility test in the adjacent teeth is negative and the vitality test shows 11 tooth is non-vital.

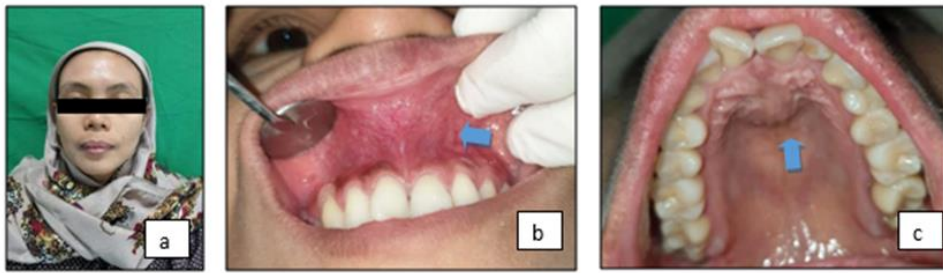


Figure 1. Frontal View (a) Buccal Vestibulum View (b) Palatal View (c)

Radiographic examination using orthopantomography (Figure 2a) presented a radiolucent lesion with a thin radiopaque border in 12 to 22 region and expand superiorly reaching the nasal base. In the central of the radiolucency showed a radiopaque image resembling a tooth. An occlusal radiograph was also conducted to assist the better view of the lesion. Fine-needle aspiration biopsy (FNAB) revealed a hypocellular specimen with PMN, MN inflammatory cells, plasma cells, and cyst macrophage. The background was wide necrotic debris. There was no malignant cell in this specimen. Thus, it could be concluded as an inflammatory, benign cystic lesion.

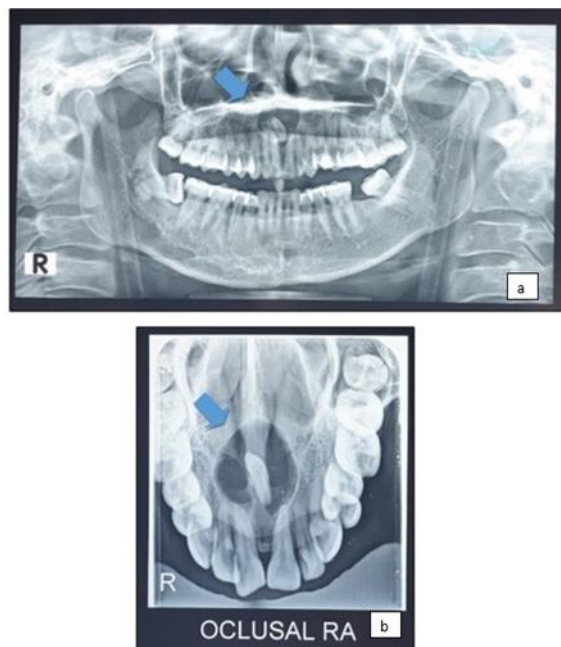


Figure 2. Orthopantomogram Radiographic Imaging (a) Occlusal Radiographic Imaging (b)

This case was treated with cyst enucleation through access from the labial side. The labial approach was taken considering upon palpation, the labial side revealed a cystic consistency compared to the hard palatal side, and to obtain a wide area of operation, a labial access was made, with a marginal incision on the gingival cervical of 13-23, continued with periosteal flap elevation and the cortical

bone was drilled until an approach to the cystic lesion was possible. Upon achieving the cyst lesion, a yellowish-brown liquid was obtained, next the cyst capsule was enucleated by separating it with the periosteum on the palatal mucosa, continued by extraction of mesiodens. Sutures were placed for defect closure. Root canal treatment on tooth 11 was scheduled on the next control visit.



Figure 3. Cyst space after enucleation (a) Dentigerous cyst lesion and mesiodens tooth (b) HPA imaging (c)

Post-surgery histopathological assessment presented a cyst wall consisted of loose fibrous connective tissues lined by squamous epithelium along with erosions, lymphocyte inflammatory cell infiltration, erythrocyte extravasation, cholesterol cleft, and vasodilation (Figure 3c). There was no sign of malignancy. Thus the lesion could be diagnosed as a dentigerous cyst.

Discussion

A dentigerous cyst is an odontogenic developmental cyst associated with the crown of unerupted teeth and developing teeth (Martinelli-Kläy et al., 2019). The prevalence of dentigerous cyst is 16.6% of all jaw cysts, around 95% of this cyst involves permanent dentition, and only 5% is involved with supernumerary teeth. Mesiodens is the most common supernumerary teeth and are often located in the midline between the central maxillary incisors (Hassnain et al., 2017). The dentigerous cyst is often asymptomatic and undetectable unless the patient undergoes screening with the orthopantomogram. This cyst can also cause pain if infected and could grow larger, causing intraoral swelling, at some times extraorally, could shift the position of teeth involved, and cause resorption of the apex of involved teeth (Carrera et al., 2013).

According to Passi D, dentigerous cyst in mesiodens could develop with liquid accumulation between the reduced enamel epithelium with the mesiodens enamel, or between the layers of the reduced enamel epithelium. Cysts occur due to the pressure from mesiodens which prevents venous flow, causing rapid serum transudation to the capillary walls (Mishra et al., 2017). The liquid filling the inner part of this cyst is hyperosmotic as a result of albumins, immunoglobulins,

and rest of squamous epitheliums. This liquid causes extracellular liquid to flow into the cyst and expansion of the cyst to surrounding tissues. The epithelial layer of the cyst then secretes factors activating collagenase and osteoclast thus causing surrounding bone resorption and increasing the size of the cyst (Widhianingrum et al., 2016).

Radiographically, dentigerous cyst is commonly seen as clear bordered, radiolucent, unilocular, and usually symmetric, surrounding an impacted crown of tooth. This cyst is retentive to the cemento-enamel junction, a feature to be pointed out in diagnosis. The inner part of the cyst is radiolucent, except for the involved crown. The hyperplastic follicle condition could further complicate distinction with differential diagnoses (Gendviliene et al., 2017). Histologically the dentigerous cyst is presented with 2-4 layers of cystic lining containing non-keratinized stratified squamous epithelium and chronic inflammatory cells on fibrous tissues containing odontogenic epithelial rests. Retepegs are uncommon except in secondary infection cases. Cholesterol clefts could also occur. The fibrous tissue capsule contains immature fibroblasts separated by mucopolysaccharide ground substance (Mishra et al., 2017).

A pericoronal radiolucency defined as a lesion diameter of more than 4 millimeters, and histologically, connective tissue surrounded by non-keratinized stratified squamous epithelium. Upon surgery, a cystic space occurs between the enamel and surrounding tissues. Upon the three criteria above, the third criteria is the most important, but all the criteria must be fulfilled to obtain a definitive diagnosis (Kolokythas & Miloro, 2015). In this case, the patient felt a lump in her palate and labial gingiva, with pain and swelling indicating inflammation on the cyst lump, and the pressure of the cyst caused rotation of (Chhabra et al., 2020), this caused loosening of the periodontal ligament and access for bacterias to enter the micro space between the teeth and the alveolar bone thus causing infection. This chronic infection could spread to surrounding bones, and the thin alveolar bone around the cyst wall makes it easier for bacteria to penetrate and causes infection to develop around the cyst (Widhianingrum et al., 2016).

Orthopantomogram imaging revealed a radiolucent lesion with a clear thin radiopaque border, the lesion expanding from regions 12 to 22 with expansion to the superior until the nasal base, approximately 2 cm in size, and also a radiopaque imaging resembling a mesiodens, with no permanent teeth root resorptions around the lesion. Upon surgery, a yellowish-brown cystic space between the enamel and its surrounding tissues was obtained. This is caused by cystic liquid mixed with pus because of the infection process. The cyst specimen was sent to the Pathology Anatomy Laboratory for HPA examination and revealed a cyst wall tissue sample covered by squamous epithelium cells with a few erosion parts. Consisted of loose fibrous connective tissues, with lymphocyte inflammatory cells infiltration and erythrocyte extravasation. Cholesterin cleft and vascular widening was also seen. No signs of malignancy occurred. From the clinical signs upon surgery, the radiologic and histopathology based on Daley and Wysoccki strengthened the diagnosis of dentigerous cyst.

The treatment option of dentigerous cyst is associated with size, location, and the possibility of recurrence of the cyst. Marsupialization is the choice of treatment

for cysts larger than 2.5 centimeters in diameter and in proximity to vital structures, thus often a preliminary method before enucleation to minimize bone defect. Cyst enucleation has been the most effective method for cyst treatment with cyst diameter less than 2.5 centimeters on the orthopantomogram because enucleation could eliminate all cyst capsule tissues, thus minimizing the recurrence of the cyst (Chhabra et al., 2020).

In this case, total enucleation of cyst along with tooth extraction was the treatment of choice, with consideration of the cyst size less than 2 cm, and located at a distance with vital structures, as well as providing an adequate access. was planned for root canal treatment upon the next visit because of involvement with the cyst lesion making it non-vital. The disadvantage of cyst enucleation is that if the enucleation was performed inadequately, it could trigger intracystic component breakage causing recurrence (Chhabra et al., 2020). The evaluation of the patient will be conducted periodically and orthopantomogram radiographic imaging on the sixth and twelfth month after enucleation would be made for evaluation of bone defect closure and bone density. This is based upon a previous study that conducted bone density evaluation periodically with the intervals of 6, 12, and 24 months after cyst enucleation . It revealed bone defect reduction gradually and a significant increase in bone density.12 Wagdargi et al, discovered that the increase of bone density was significant (90.8%) 6 months after enucleation and there was primary defect closure in 16 odontogenic cysts with sizes ranging from 3 to 10 centimeters, and an increase in bone density in mandibular cavities compared to the maxillary cavities (Garg et al., 2016).

The prognosis of dentigerous cyst is excellent, with infrequent recurrence. A study revealed that dentigerous cyst does not recur after complete excision. A longitudinal study reported the management of dentigerous cyst for 11 years, with 40 comprehensive dentigerous cyst cases (involving more than three teeth) on the maxilla and mandibula, with no recurrence in all of the cases (Garg et al., 2016). Even though it has a good prognosis, an untreated dentigerous cyst could grow and cause pathological jaw fracture and could develop ameloblastoma, squamous cell carcinoma, and mucoepidermoid carcinoma (Kondamari et al., 2018).

Conclusion

It could be concluded that two abnormalities such as dentigerous cyst in the maxilla and its relation with mesiodens is a rare case. Dentigerous cyst management considers its location, size, and cyst enucleation is the most effective method for cyst treatment. Mesiodens is best diagnosed as early as possible and treated promptly to prevent dentigerous cyst destructing the teeth and vital structures around it.

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