Case Report

Dentigerous Cyst Crossing The Midline Associated With Multiple Inverted Supernumerary Teeth And Impacted Permanent Tooth: A Case Report.

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ABSTRACT

Dentigerous cysts are the second most common odontogenic cysts after radicular cysts and are most commonly seen in association with the third molar region of the mandible, followed in frequency by maxillary canine, maxillary third molar, and rarely in relation to maxillary central incisor. Dentigerous cysts are the most common developmental cysts of the jaws. Dentigerous cysts around supernumerary teeth, however, account for 5% of all dentigerous cysts, with most developing around a mesiodens in the anterior maxilla. This article reports a rare and unusual case of a dentigerous cyst of the anterior maxilla involving a multiple inverted mesiodens with permanent impacted tooth with midline crossing. The complications associated with inverted mesiodens include eruption disturbance of adjacent teeth, displacement & rotation of the central incisors, diastema, root resorption, abnormal occlusion, cyst formation or nasal eruption of the mesiodens. Early detection and timely surgical intervention of inverted mesiodens is crucial to prevent unwanted consequences. A provisional diagnosis of infected odontogenic cyst was made. Histopathological examination confirmed the diagnosis of infected dentigerous cyst. The patient was treated surgically by enucleation of total cyst and surgical extraction of multiple inverted mesiodens and permanent central incisor under general anesthesia.

Key Words: dentigerous cyst, inverted tooth , midline crossing, multiple supernumerary tooth

INTRODUCTION

The dentigerous cyst is defined as a cyst that originates by the separation of the follicle from around the crown of an unerupted tooth.[1] Dental follicle associated with unerupted or impacted teeth shows fibrous connective tissue with remnants of reduced enamel epithelium. Dentigerous cysts are caused by expansion of dental follicles resulting from accumulation of fluid between tooth crowns and epithelial components.[2] The Dentigerous cyst is the most common type of non-inflammatory odontogenic cyst and the most common cause of a pericoronal area of lucency associated with an impacted tooth.[3]

Dentigerous cysts are the second most common odontogenic cysts after radicular cysts, accounting for approximately 24% of all true cysts in the jaws. [4]This cyst most frequently occurs in patients between 10 and 30 years of ages and there is a greater incidence in males with a 1.6:1 ratio. [1,5] Stafne first described dentigerous cysts associated with supernumerary teeth and found an incidence of 5.5% among 200 supernumerary teeth.[6] Dentigerous cysts occur predominantly in the third molar region of the mandible, followed in frequency by maxillary canine, maxillary third molar, and rarely in relation to maxillary central incisor.[7]

They are occasionally associated with supernumerary teeth [8,9]Their association with supernumerary teeth was studied by Lustmann and Bodner in 1988.[10]

A mesiodens, a kind of supernumerary tooth, is located in the maxillary central incisor region. A mesiodens has an overall prevalence of 0.15-1.9%.

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A mesiodens can occur individually or in multiples and often does not erupt. Mesiodens sometimes bring about median diastema, delayed eruption of the permanent central incisors, alteration of the position of the permanent incisors, and dentigerous cyst formation.[11]Most supernumerary teeth are noted in the anterior maxillary region. The most common supernumerary tooth which appears in the maxillary midline has also been named a mesiodens due to its position in the center of the maxilla [12]

Histologically, a dentigerous cyst is lined by non-keratinized stratified squamous epithelium. Since the dentigerous cyst develops from follicular epithelium it has more potential for growth, differentiation and degeneration than a radicular cyst. Occasionally, the wall of a dentigerous cyst may give rise to a more ominous mucoepidermoid carcinoma. Due to the tendency for dentigerous cysts to expand rapidly, they may cause pathological fractures of jaw bones.[13]

The usual radiographic appearance is that of a well-demarcated radiolucent lesion attached at an acute angle to the cervical area of an unerupted tooth. The border of the lesion may be radiopaque. The radiographic differentiation between a dentigerous cyst and a normal dental follicle is based merely on size. Radiographically, a dentigerous cyst should always be differentiated from a normal dental follicle. Dentigerous cysts are the most common cysts with this radiographic appearance. Radiographically the cyst appears unilocular with well-defined margins and often sclerotic boarders. Infected cysts show ill-defined margins.[13]

It may cause facial asymmetry if aggressive in nature.[3]

Dentigerous cysts by far the most common lesion that often exhibits marked displacement of the unerupted tooth within the jaw. With the pressure of enlarging cyst, the unerupted tooth can be pushed away from its direction of eruption.[14]

Here, we present a rare case report of a Dentigerous cyst in an 27-year-old male in the anterior mandible crossing the midline associated with multiple inverted supernumerary teeth along with impacted underdeveloped permanent central incisor. This rare case is reported because of its unusual presentation at such a location, and most importantly it is unique case because it is a single Dentigerous cyst crossing midline, which is reported in only few literature available.

CASE REPORT

A 27-year-old male patient reported to the Department of stomatology at The first affiliated hospital of Yangtze university, jingzhou, china with a chief complaint of painful swelling in the maxillary right vestibular region for 6 months. Since then, the swelling had gradually increased in size. There was no swelling in the hard palate. In maxillary dentition right canine was rotated and maxillary left central incisor tooth was missing. There was no history of trauma or no dental treatment around the lesion. The patient's neurological exam was normal. The radiograph of the lesion revealed an impacted two supernumerary teeth with a maxillary left central incisor tooth which was associated with a well-defined radiolucent area in the anterior region of the maxilla.(fig 1)

In routine blood investigations blood sugar was found markedly elevated and thus patient was diagnosed as diabetes mellitus by endocrinology department. There was no history of allergic symptoms. The patient had taken an unknown medication for the pain and swelling, but he had no relief.

On clinical examination, a solitary diffuse extra-oral swelling was seen in the upper right maxillary region of the face. It had extended from left first upper premolar to right upper first molar crossing the midline. The tip of the nose was slightly deviated towards the left side, and the right naso-labial fold was elevated.(fig 2)

On intraoral examination, a solitary well-defined intraoral swelling measuring about 10cm×3 cm was seen in the maxillary anterior labial vestibule extending from the left first premolar up to the right first molar. The swelling was roughly oval in shape. The swelling was tender on palpation and fluctuant in consistency. The overlying mucosa was bluish in color, and the superficial blood vessels were prominent.

The panoramic radiograph showed a well-circumscribed, corticated radiolucent lesion in the anterior maxilla, crossing the midline and extending from the left maxillary first premolar to the right maxillary second molar on the mesial aspect. The root of maxillary right canine, lateral incisor, central incisor and left maxillary lateral incisor are slightly displaced.(fig 1)

On a computed tomography (CT) scan, the coronal and axial section showed a large, radiolucent, well-defined

lesion in the anterior maxilla with a two inverted supernumerary teeth and one impacted permanent maxillary left central incisor seen towards the inferior border of the lesion involving right maxillary sinus .The two supernumerary teeth and one impacted tooth was on labial aspect of maxilla. There was upward displacement of the floor of the nasal cavity.

A provisional diagnosis of the infected cyst was made on the basis of the patient's history, clinical examination, and radiographic examination. On aspiration, a viscous brick-red colored fluid was suggestive of an infectious cyst.

The patient was taken up for surgery under general anesthesia after the health condition of patient was normal. The cyst was approached by a crevicular incision with three corner flap extending from maxillary left second premolar to maxillary right second molar.(fig 4) The outer cortex was found to be paper thin. The bluish cyst was identified and excised. The lesion was surgically enucleated along with removal of the impacted two inverted supernumerary teeth and one underdeveloped maxillary left central incisor(fig 5) To control the bleeding from cystic cavity we placed a balloon catheter prepared by the hand surgical glove and rubber drainage tube which was passed through inferior nasal miatus into the cystic cavity.(fig 6) The excised specimen showed two supernumerary teeth and one maxillary left central incisor without formation of the root, which are deeply embedded in the cyst

The histopathological examination showed a cystic lumen with a very thin lining of stratified squamous epithelium and dense fibro cellular connective tissue stroma with chronic inflammatory infiltrate, suggestive of an infected dentigerous cyst.

The patient remained asymptomatic, and no complications were noted after 6 months follow-up.

Fig 1: OPG showing a unilocular well defined radiolucency with scalloped margins associated with two impacted supernumerary teeth and one impacted permanent maxillary central incisor.

Fig 2: Clinical photograph shows a solitary diffuse extraoral swelling in the upper anterior region of the face. The tip of the nose is slightly deviated towards the left side, and the right naso-labial fold is elevated.

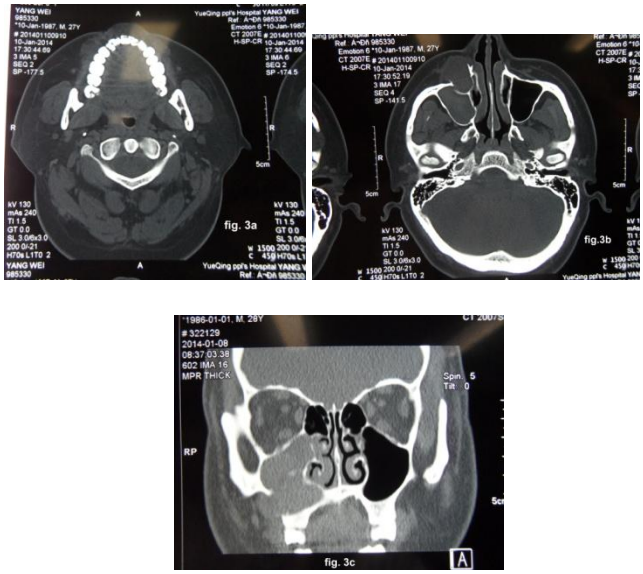

Fig 3: CT fig 3a- axial, fig 3b- axial and fig 3c- coronal view reveals cystic cavity involving right maxillary sinus crossing mid line associated with supernumerary teeth.

Fig 4: After excision of the cystic lining

Fig 5: Tissues specimen with 2 supernumerary teeth and one permanent tooth.

DISCUSSION

The term dentigerous is preferred, the literal meaning being "tooth bearing."[15]The Dentigerous cyst occurs most commonly in second and third decade [16] Koseoglu et al. in their study, stated that Dentigerous cysts occur over a wide age range age from 15 years to 65 years.[17] As evident from literature Dentigerous cyst is twice as often in males than females and is 10 times more likely to occur in the lower-jaw than in the upper-jaw. The present case seems to support the age and sex pre-dilection given by other authors [18]. A study reported by Mourshed state that 1.44 per cent of impacted teeth may undergo Dentigerous cyst transformation.[19]

A mesiodens is known to have a cone-shaped crown and a short root, as seen in our patients. It is a rare entity with a reported incidence of 0.15% to 1.9%, and it has a slight predominance in males [20]. Primosch reported an enlarged follicular sac in 30% of the cases, but the histological evidence of cyst formation was found in only 4-9% of the cases [21].

The direction of eruption of a mesiodens can be divided into 3 groups, viz. normal, inverted or horizontal direction. Most common ocuurence of mesiodens reported in literature have been the invertred type, which were impacted in most of the cases. [22-25] Mesiodens can occur singly or in multiples is one of the responsible factors for the disturbances in the eruption of maxillary incisors. [26-27]. Most mesiodens never erupt and usually found to be impacted, with a conical crown and a single root, and often in an inverted position [28-29] as in our case. But when they do erupt, the most common site is behind the central incisors within the premaxilla. [26]

Inverted eruption of teeth has been defined as the 'malposition of a tooth in which it has reversed and is positioned upside down'. Inverted teeth have been reported in both maxilla and mandible, and most of them are inverted impacted third molars and premolars. [29]

One of the common developmental odontogenic cysts, viz. the dentigerous cyst usually is detected on routine radiographic examinations. While a developing dentigerous cyst is difficult to differentiate from a normal follicle, any pericoronal radiolucencies more than 4 mm should be considered cystic, unless proven otherwise [30].A radiograph of a patient with dentigerous cyst reveals a unilocular radiolucency enclosing the crown of an unerupted tooth. The radiolucency usually arises in the cemento-enamel junction of the tooth [29]. If a follicular space on the radiograph appears to be more than 5 mm, then presence of an odontogenic cyst can be suspected. Differential diagnoses of such radiolucencies include radicular cyst, odontogenic keratocyst, and odontogenic tumors such as ameloblastoma, Pindborg tumor, odontoma, odontogenic fibroma, and cementomas.[31]

Of the 95% dentigerous cysts which involve the permanent dentition only 5% are associated with supernumerary teeth.[26] Studies have shown that about 6% of supernumerary teeth may develop dentigerous cyst.[28] .The cysts most often involve impacted mandibular third molars, followed by maxillary canines, mandibular premolars, and occasionally supernumerary teeth or odontomas.[22,32]

Dentigerous cyst involving an impacted inverted mesiodens has been reported that developed secondary to trauma to permanent maxillary central incisors. The root development was arrested prematurely with the open apex lying in close proximity to the underlying inverted mesiodens.[33]

Clinical diagnosis may be confused with other types of odontogenic cysts. Histopathology of the excised specimen confirms the diagnosis. Management of a supernumerary tooth usually warrants its removal, especially when associated with definite pathology.[29]

There is no evidence of Dentigerous cyst crossing the midline in the literature in various search engines like

Medline, Cochrane and PubMed apart from a case report by Gonzalez et al. (2011) who reported a Dentigerous cyst encompassing the right and left impacted mandibular canines and crossing the midline. Hence, we are reporting this case, which is unique in its own kind as Dentigerous cyst has never been found to cross midline. This will be a great break-through in the classification of midline crossing cysts. The authors also stated that as Dentigerous cyst enlarges, it displaces the affected tooth or teeth apically, which was found in accordance to our case report.[34]

Many dentigerous cysts are small asymptomatic lesions that are discovered serendipitously on routine radiographs, although some may grow to considerable size causing bony expansion that is usually painless until secondary infection occurs. Since cysts can attain considerable size with minimal or no symptoms, early detection and removal of the cysts is important to reduce morbidity. Moreover, almost all of the reported cases, present without pain and discovered during the investigation of asymptomatic slowly-growing swellings. Radiographically, the dentigerous cyst presents as a well-defined unilocular radiolucency, often with a sclerotic border. Since the epithelial lining is derived from the reduced enamel epithelium, this radiolucency typically and preferentially surrounds the crown of the tooth. A large dentigerous cyst may give the impression of a multilocular process because of the persistence of bone trabeculae within the radiolucency. However, dentigerous cysts are grossly and histopathologically unilocular processes and probably are never truly multilocular lesions.[13]

Three types of dentigerous cyst have been described radiographically: The central variety, in which the radiolucency surrounds just the crown of the tooth, with the crown projecting into the cyst lumen. In the lateral variety, the cyst develops laterally along the tooth root and partially surrounds the crown, the circumferential variant exists when the cyst surrounds the crown but also extends down along the root surface as if the entire tooth is located within the cyst. Our case was radiographically a classic presentation of the circumferential variety.

The histological features of dentigerous cysts may vary greatly depending mainly on whether or not the cyst is inflamed. In the non-inflamed dentigerous cyst, a thin epithelial lining may be present with the fibrous connective tissue wall loosely arranged with inflammatory cells. As the lining is derived from reduced enamel epithelium it is 2-4 cell layer thick primitive type. The cells are cuboidal or low columnar. Retepegs formation is absent except in cases that are secondarily infected. As the connective tissue wall is derived from the dental follicle of developing enamel organ, it is a loose connective tissue stroma, which is rich in acid mucopolysaccharides. In the inflamed dentigerous cyst, the epithelium commonly demonstrates hyperplastic rete ridges and the fibrous cyst wall shows an inflammatory infiltrate. The content of the cystic lumen is usually thin watery yellow fluid and is occasionally blood tinged. Histopathogenesis of dentigerous cyst is based on intra-follicular and extra-follicular theories. There can be no good reason for the extra-follicular theory of origin of dentigerous cysts as the evidence is that those reported as arising in this manner all appear to been envelopmental or follicular odontogenic keratocyst.[13] Intra-follicular theory postulates the possibility of cyst formation due to fluid accumulation between the layers of inner and outer enamel epithelium after the formation of crown. Main's theory.[35]The impacted tooth exerts pressure on the follicle which obstructs the venous outflow and induces rapid transudation of serum across the capillary walls. The increased hydrostatic pressure exerted by this pooling of fluid causes separation of the crown from the follicle with or without reduced enamel epithelium. The osmolality of the cyst fluid is modified by increased permeability to glycosaminoglycans such as hyaluronic acid, heparin and chondroitin sulphate, which causes expansile growth rapid.[36]

Most dentigerous cysts are treated with enucleation of the cyst and removal of the associated tooth. Large dentigerous cysts may be treated with marsupialization when enucleation and curettage might otherwise result in neurosensory dysfunction or pre-dispose the patient to an increased chance of pathological fracture. Occasionally, it transforms to squamous cell carcinoma, mucoepidermoid carcinoma or ameloblastoma from or in association with a dentigerous cyst.[37-39].The prognosis for most histopathologically diagnosed dentigerous cysts is excellent, recurrence being a rare finding.[13,40]

Water's, panoramic, and skull radiography are simple and inexpensive methods that can be used in daily practice. The structure of a tooth can be clearly detected on panoramic radiographs. Therefore,

panoramic radiographs are preferred over CT.[41] Although the structure of a tooth can be clearly detected on panoramic radiographs, they are inadequate for localizing maxillary ectopic teeth due to their inherently less sharp image and ghost image. CT scan provides superior bony detail, allowing for the visualization of the size and extent of the lesion with determination of orbital or nasal invasion or involvement.[4] Therefore, CT may be more valuable than plain film radiographs, not only for definitive diagnosis, but also for evaluation of the associated pathology, exact localization of the ectopic tooth, and proper treatment planning.[42]

In our case, dentigerous cyst was treated by the enucleation and removal of associated impacted teeth. With the placement of balloon catheter on cystic cavity through inferior nasal meatus to control postoperative bleeding and future reduce the secondary infection.

CONCLUSION

This new case presentation shows that a dentigerous cyst might also cross the midline. As dentigerous cyst may be symptomatic or asymptomatic they can attain considerable size without the notice of the patient and this warrants an early clinical and radiographic detection of the cyst so that early treatment strategies will prevent or decrease the morbidity associated with the same. So if any permanent tooth is missing or any crowding of teeth is present we have to consider for further radiographic investigations. Therefore, supernumerary teeth or impacted teeth should be examined carefully to prevent harmful effects on the adjacent regular teeth and possible cystic development.

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