CASE REPORT

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A large radiolucent lesion with impacted teeth: was it an ameloblastoma or a dentigerous cyst? a case report

Galih Rahmadini[1,2*], Belly Sam[3], Suhardjo Sitam[3], Azhari[3], Seto Adiantoro Sadputranto[4]

ABSTRACT

Objectives: This case report aims to describe a large radiolucent lesion associated with impacted teeth from CBCT radiographs.

Case Report: A 19-year-old woman was referred to have a CBCT examination in the dentomaxillofacial radiology unit in Dental Hospital Universitas Padjadjaran with swelling on the palatal side of the face and a lump on the gum at the upper right unerupted canine and premolar. The CBCT examination results showed impacted teeth 13 and 14 with a large radiolucent lesion with a well-defined and corticated border surrounding teeth 13 and 14, expanding into the maxillary sinus and nasal cavity. Oral hygiene was in good condition. The facial profile looked asymmetrical.

Conclusion: Based on the CBCT result in terms of location and radiographic feature, this extensive radiolucent lesion led to a suspect radiodiagnosis of the ameloblastoma. Histology examinations are required to establish a definitive diagnosis. A cyst wall lined with odontogenic squamous epithelium was confirmed in the histology result examination.

Keywords: *Ameloblastoma, CBCT, dentigerous cyst, impacted teeth, large radiolucent*
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INTRODUCTION

The term "impacted tooth" refers to a situation in which a tooth will probably not erupt spontaneously into the oral cavity at the estimated time based on dental age without the aid of orthodontic treatment and or surgical intervention. The incidence of impacted teeth ranges from 0.8% to 3.6% of the population, with wisdom teeth, maxillary canines, mandibular second premolars, and maxillary lateral incisions being the most common teeth.[1] An impacted tooth is defined as a tooth that fails to erupt after the normal development pattern is complete, while a normal tooth erupts when half to three-quarters of its final root length has developed. The etiology of tooth impaction is multifactorial. In the literature, the most commonly reported etiological factors related to tooth impaction can be divided into three different groups: systemic, local, and genetic.[2] Delayed tooth eruption or the presence of impacted teeth can cause fluid accumulation in the epithelial layer of tooth enamel. When impacted teeth are retained within the alveolar process, the associated follicular sac is also retained.

Although in most patients the dental follicle maintains its original size, it may undergo cystic degeneration and become a dentigerous cyst or keratocyst. If the patient is closely followed, the dentist can diagnose the cyst before it reaches large proportions.However, unmonitored cysts can reach enormous sizes. As a general guideline, if the follicular space around the crown of the tooth is greater than 3 mm, the diagnosis of a dentigerous cyst is a reasonable one.[3] A dentigerous cyst form around and covers the crown of an unerupted tooth, attached from the crown to the upper root within the cemento-enamel junction (CEJ). This cyst may be cause by a developmental abnormality or inflammatory.[4] They usually present in the second and fourth decades of life but are uncommon in childhood as they exclusively occur in the secondary dentition. They're also known as follicular cysts which is normal development of the tooth. The incidence of dentigerous cysts has been reported as 1.44 in every 100 unerupted teeth.[5] Dentigerous cysts constitute of 20% from all odontogenic cyst 20% of all odontogenic cysts, developing during during 10 to 30 years, with the most predisposition to male (3:2).[6]

A dentigerous cyst has a differential diagnosis feature with ameloblastoma because of the similarity of the radiographic appearance, may be a well-defined unilocular radiolucency with sclerotic borders, multilocular radiolucencies with scalloped borders and involving teeth impaction.[7] An ameloblastoma is a benign and a locally aggressive tumour which arises from the mandible or less commonly, from the maxilla. Unicystic ameloblastomas are variants of ameloblastomas,

[1]Dentomaxillofacial Radiology Residency Program, Faculty of Dentistry, Universitas Padjadjaran, Bandung, Indonesia 40132

[2]Cijulang Pangandaran Health Center, Pangandaran, Indonesia 46394

[3]Department of Dentomaxillofacial Radiology, Faculty of Dentistry, Universitas Padjadjaran, Bandung, Indonesia 40132

[4]Department of Oral and Maxillofacial Surgery, Hasan Sadikin Central General Hospital, Bandung, Indonesia 40161

*Correspondence to:
Galih Rahmadini
galih21004@mail.unpad.ac.id

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which were first described by Robinson and Martinez, which refer to those cystic lesions that show clinical and radiological characteristics of odontogenic cysts, but which on histological examination, show typical ameloblastomatous epithelium which lines part of the cyst cavity, with or without a luminal or mural tumour proliferation.[8]

Fifteen to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts. Since 1925, many had reported the development of ameloblastomas within the walls of odontogenic cysts, among which the most commonly cited were dentigerous cysts. When the ameloblastoma is associated with retained teeth, unicystic ameloblastomas share many clinical and radiological similarities with dentigerous cysts. This similarity is so strong that, many times, it is possible to differentiate them just by means of a histopathological examination.

A dentigerous cyst and ameloblastoma has the propensity to displace and resorb adjacent teeth. The associated tooth is frequently displaced in an apical direction. There may be a significant amount of displacement. Third molars in the maxilla or cuspids can be moved to the floor of the orbit, and third molars in the mandible can be moved to the condylar, coronoid, or inferior cortex of the mandible. The floor of the maxillary antrum may be displaced as the cyst invaginates the antrum, or a cyst may displace the inferior alveolar nerve canal in an inferior direction. This slow-growing cyst often expands the outer cortical boundary of the involved jaw. This cyst that invaginates the sinus can drain and collapse with new bone formation at the periphery.[9]

Radiographic evaluation is an important diagnostic procedure necessary to (a) detect the extent of large lesions, (b) identify anatomical relationships, and (c) assess the margins of the lesion to ascertain the presence of infiltrative growth suggestive of development of an aggressive lesion.[10] Dentigerous cyst dan ameloblastoma in the maxillary sinus are not difficult to identify radiographically because of their opacity. In these instances, panoramic radiography is an appropriate technique for determining the presence of this lesion. However, Computed Tomography (CT) scan can also assist in determining the precise size or content of lesions that extend into the maxillary sinus and identifying bony structures. Cone Beam Computerized Tomography (CBCT) radiographic examination is one of the supporting examination options to diagnose this case. This modality offers wide three-dimensional imaging of a mineralized view of maxillofacial tissue, with minimum distortion and a much lower radiation dose than conventional CT. CBCT is the right method to assist diagnosis, this radiographic technique provides greater visualization from multiplanar which accommodates the user to detect the abnormality with its surrounding associating anatomy.[11]

This paper illustrates a case of an unusual dentigerous cyst initially diagnosed as ameloblastoma due to its multiloculated feature observed in CBCT.

CASE REPORT

A 19-year-old woman was referred to have a CBCT examination in the dentomaxillofacial radiology unit in Universitas Padjadjaran Dental Hospital with swelling on the palatal side of the face with a lump on the gum at the upper right unerupted canine and premolar. Facial profile looked asymmetrical (Figure 1), oral hygiene was in good condition (Figure 2) and panoramic radiography was taken (Figure 3). From the subjective and objective examination, a biopsy was taken and the patient was reffered for CBCT radiograph examination. The CBCT examination (Figure 4 and 5) results showed impacted tooth 13 and 14 with a large radiolucent lesion with clear, well-defined and corticated border around of tooth 13 and 14. The lesion extends superiorly, destroyed the cortical floor of the maxillary sinus and nasal fossa. Although this lesion does not resorb the root of the tooth, it could be seen this lesion makes an expansion of the maxillary dextra bone, displacement of tooth 13 and tooth 14, get into the sinus until the right sinus cavity is filled with fluid.

CBCT revealed a huge well-defined radiolucency unilocular and fill the maxillary sinus. The periphery of the lesion is well defined, not surrounded by any sclerotic border. The extension of the lesion

Figure 1. Extraoral examination revealed a diffuse swelling over the right side of the face

Figure 2. Intraoral examination revealed a diffuse swelling in the palatal site from 14 to the posterior region

Figure 3. Panoramic radiographic examination, showing the impacted teeth 13 and 14 with the involvement of the maxillary sinus and nasal fossa. Note the presence of unilocular lesion around the vertically impacted tooth 13 and horizontally impacted 14.

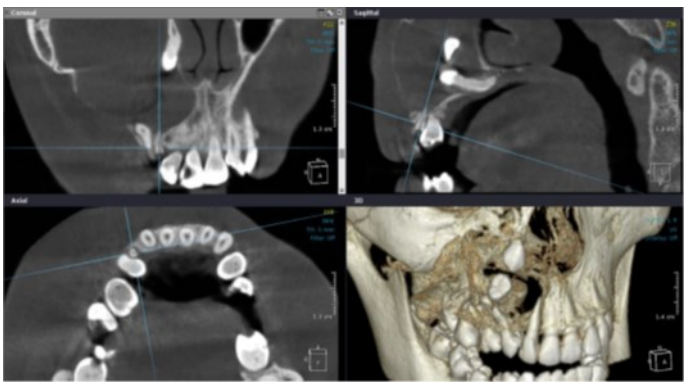

Figure 4. Multiplanar (MPR) view of the impacted teeth with a large radiolucent lesion on the right maxilla

superiorly depresses tooth 13 into the nasal fossa and the nasal fossa also depressed by the lesion. While the internal density of the lesion indicates a fluid/fatty tissue, there was discontinuity in the floor of the right maxillary sinus and suspected that the cyst fluid can come out of the lesion and enter the sinus. Based on the shape and pattern of the lesion from the results of the CBCT examination, the radiodiagnosis of this case was unicystic ameloblastoma with differential diagnosis of dentigerous cyst.

Incisional biopsy was performed and sent for histopathological analysis. The maxillary dextra biopsy specimens in the form of pieces of tissue from the cyst wall covered with odontogenic squamous epithelium which is partially erosive, the core is within normal limits, the subepithelial consists of fibrocollagenous connective tissue stroma covered with inflammatory cells, lymphocytes, PMN cells, histocytes and bleeding. Also visible bone structure with osteocyt cells within normal limits with visible osteoblast cells at the edges. The nucleus is within normal limits and there is no sign of malignancy. Based on the results of histological examination, a diagnosis of dentigerous cyst was made.

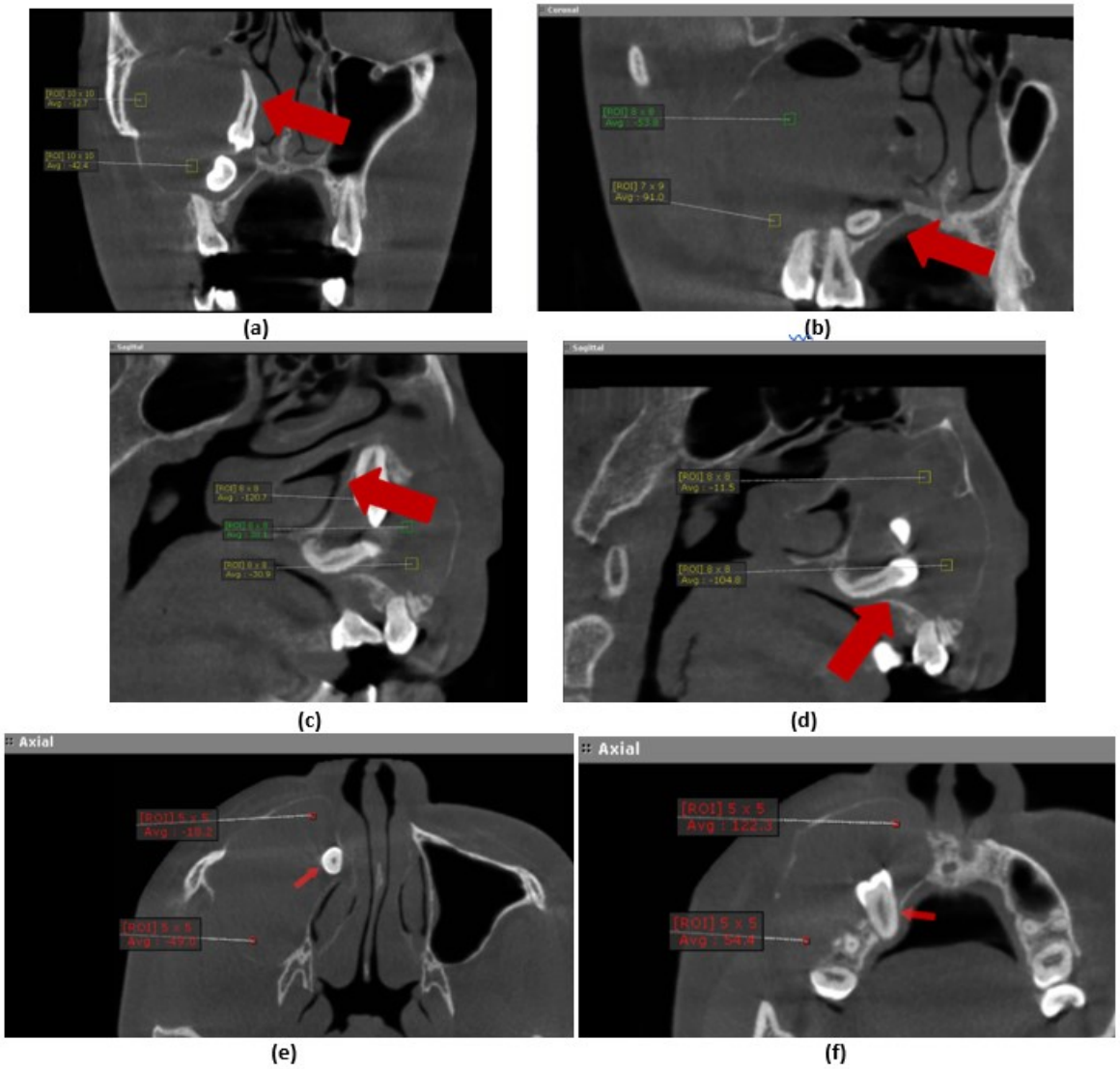

Figure 5. Coronal, sagittal and axial slicing view of CBCT radiograph. (a,c,e) There is a radiolucent lesion with well corticated on the right anterior maxilla that extends to the maxillary sinus, destroying the cortical floor of the maxillary sinus. The position of the impacted tooth 13 is vertical with the mesial ¾ of the tooth perforated into the nasal fossa. A radiopaque image appears resembling the tooth seed 14 which is impacted above the incisal tooth 13 (b,d,f) The position of the impacted tooth 14 is horizontal with the root lacerated towards the nasal fossa and the apical half of the tooth destroying the alveolar bone.A radiolucent well corticated around the impacted tooth is seen.

DISCUSSION

The order of frequency of dental impactions begins with the 3rd molars, followed by maxillary canines, mandibular premolars, mandibular canines, maxillary premolars, maxillary incisors and, finally, lower 2nd molars (lower incisors and 1st and 2nd molars jaws very rarely suffer from this anomaly). Impaction of maxillary canines is a frequently encountered clinical problem. Maxillary canines are the second-most frequently impacted teeth after the third molars with prevalence from 0.8-5.2 % depending on the population examined. The incidence of maxillary canine impaction is about 20 times more than mandibular canine impaction. The cause of canine impaction can be the result of localized, systemic or genetic factor(s). [12] Several local factors such as tooth size– arch length discrepancies; failure of the primary canine root to resorb; prolonged retention or an early loss of the primary canine; ankylosis of the permanent canine; cyst or neoplasm; dilaceration of the root; absence of the maxillary lateral incisor; variation in root size of the lateral incisor (peg-shaped lateral incisor); and variation in the timing of lateral incisor root formation, are believed to play critical roles in canine impaction.[13]

The literature pertaining to impacted premolars is limited and not extensive. The prevalence of impacted premolars has been found to vary according to age. The overall prevalence in adults has been reported to be 0.5% (the range is 0.1–0.3% for maxillary premolars and 0.2–0.3% for mandibular premolars). A lack of space in the jaw, mesial drift of teeth caused by the premature loss of primary molars, abnormal or ectopic positioning of developing premolar tooth buds, inflammatory or pathological lesions like dentigerous cysts, and other factors can all contribute to impacted premolars. In these instances, environmental or

genetic factors may also be involved.[14]

in our case, we initially suspected that the patient had ameloblastoma with the characteristics of an impacted tooth, the lesion tends to cause a lot of destruction of the surrounding tissue, resulting in facial asymmetry, based on the location according to Wilbeer et al.[15] also explained that ameloblastoma is possible in the upper jaw, So, radiographically we suspected that the patient had ameloblastoma. In reality, this is still insufficient to make a diagnosis. In this case, an impacted tooth 13 and 14 was seen with a large, well-defined radiolucent lesion around the tooth located at the maxillary sinus and nasal fossa. Abnormal tooth eruption, caused by impaction, can cause dentigerous cysts.[16] Impacted teeth cause transudation of fluid across the capillary walls. Hydrostatic pressure separates the follicle from the crown, causing the cyst to expand. Expansion of this cyst is associated with epithelial proliferation, resorption factors, and increased fluid osmolality of the cyst.[17] In this case, the radiodiagnosis of ameloblastoma was confirmed as a dentigerous cyst by histopathological examination with the characteristic a cavity lined by a non-keratinized stratified epithelium containing between two and three layers of cuboidal and/or flattened cells. The connective tissue wall is usually fibrous and often devoid of inflammatory cells.[18]

The incidence of dentigerous cyst is highest in the second and third decades of life. This cyst is usually rare in the first decade. A dentigerous cyst is considered a developmental cyst by nature and rarely causes pain unless causing a problem in a nearby structure. The radiographic appearance of a dentigerous cyst is a radiolucent symmetrical, unilocular, well-defined, cortical, and surrounds the crown of an unerupted (impacted) tooth, but in this case the lesion around the teeth and tooth 13 was migrated to the nasal fossa. The slow and regular growth of the cyst makes the dentigerous cyst have a well-defined sclerotic margin, with a well-defined cortex, and It is characterized by a thin radiopaque border. The radiographic appearance of this cyst needs to be distinguished from the normal appearance of the circum-coronal follicular space surrounding the tooth to erupt. In other cases, a radiolucent area may appear lateral to the crown of the tooth, especially if the cyst is relatively large in size or if there has been a change in the position of the tooth from its place.[16] The dentigerous cyst has the potential of expanding causing medullary bone destruction and jaw enlargement. The dentigerous cyst tends to push and resorb the adjacent teeth. The cyst generally develops in one tooth but could also involve a few surrounding teeth if the cyst grows larger. This then might cause a shift of the tooth far from its normal position especially the cysts that occur in the upper jaw teeth. The shift of the impaction and the dentigerous cyst itself could cause disruption towards the surrounding tissue. When the cyst in the maxillary sinus becomes symptomatic, the patient would experience sinusitis symptoms including swelling, facial pain, headache, and nasolacrimal duct obstruction.[17]

Radiologically differential diagnosis of dentigerous cyst is mainly made with hyperplastic dental follicle, odontogenic keratocyst, and unicystic ameloblastoma. Few cases reported in literature show the neoplastic potential of the epithelium of dentigerous cyst to ameloblastoma, epidermoid carcinoma, and mucoepidermoid carcinoma. An ameloblastoma is a locally aggressive benign epithelial odontogenic tumor, commonly arising from the mandible mostly in third to fifth decades. It occurs commonly in molar-ramus region of mandible. Ameloblastoma may arise from the remnants of dental lamina and enamel organ or from the basal layer of oral epithelium as well as the epithelium of dentigerous cyst. Radiographically, it appears as unilocular or multilocular radiolucency with a typical honeycomb or soap bubble appearance. As a result of this presumption, dentigerous cysts should be managed with caution because they are preameloblastomatous.[19] In view of the reported ameloblastomatous potential of dentigerous cysts, it is thus important to be able to recognize true ameloblastomatous epithelium from ameloblastoma-like epithelium. In most cases of odontogenic cysts, the presence of an ameloblastomatous epithelial lining in inflamed odontogenic cysts is insufficient to diagnose unicystic ameloblastomas, unless other more diagnostic features of unicystic ameloblastomas are evident.[11] In such cases, other diagnostic criteria which are included to make a diagnosis of unicystic ameloblastomas, as were described by Vickers and Gorlin, are cysts which are lined by an ameloblastic epithelium, with a tall columnar basal layer, sub-nuclear vacuole, reverse polarity of hyperchromatic nucleus and a thin layer of oedematous, degenerating stellate reticulum like cells on surface.[8]

In this case, the most likely differential diagnoses were unicystic ameloblastoma. The radiologic findings that influenced the differential diagnosis were primarily the effect of the lesion on adjacent anatomic structures and unilocular contours. On the other hand, although 3D CBCT X-ray allowed us to assess the volume of the lesion and the relationship between the lesion and the surrounding structures, the diagnosis remains the same as that inferred based on panoramic.[21]

CONCLUSION

A specific cyst or tumor may be identified if a large radiolucent lesion associated with an impacted tooth is radiodiagnosed. It is critical for the radiologist, pathologists and clinicians to determine whether a dentigerous cyst is undergoing any ameloblastomatous transformation for proper diagnosis and treatment planning. Based on the CBCT finding in terms of location and radiographic featured and histology examinations are required to establish a definitive diagnosis.

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FOOTNOTES

All authors have no potential conflict of interest to declare for this article. Informed consent was obtained from the patient for being included in this case report.

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