

Cyst Associated with an Unerupted Tooth: Diagnostic Dilemma for the Clinician

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Abstract

Common pathological conditions associated with an impacted tooth are dentigerous cyst, unicystic Ameloblastoma and odontogenic keratocyst. These conditions may create dilemma for clinician regarding proper diagnosis and treatment planning. Definitive diagnosis should be based on radiographic and histopathological examination. A case report of an 8 year old boy with cystic swelling associated with unerupted right mandibular first molar was presented here.

Keywords: Cysts; Unerupted Tooth; Impacted Tooth Unicystic Ameloblastoma (UA)

Introduction

Cystic lesions that are associated with an impacted tooth mainly includes dentigerous cyst (16 - 24%), unicystic ameloblastoma (UA) (10 - 15%), keratocystic odontogenic tumor (12 - 14%), ameloblastic fibro-odontomas (1.7 - 4.6%), odontomas (22%) and adenomatoid odontogenic tumor (0.1 - 3%) [1]. Dentigerous cysts are developmental odontogenic cysts that typically show a well-defined unilocular radiolucent area around the crown of an unerupted tooth [2]. They are most common pathology associated with impacted tooth (1.44 cysts in every 100 unerupted teeth) [1]. Unicystic Ameloblastoma is a type of ameloblastomas, associated with at least one unerupted tooth (mostly the third molar) in 50% and 80% of cases [3]. Their typical appearance includes a well-defined radiolucency surrounding the crown of an unerupted tooth with bone expansion, perforation of cortical bone, and root resorption. World Health Organization (2005) defined odontogenic keratocysts as intraosseous tumors of odontogenic origin and recommended the term keratocystic odontogenic tumor for this entity [1]. Mostly they involve tooth-bearing areas and associated with at least one impacted tooth in roughly 27% of cases (mostly third mandibular molar) [1,4]. Adenomatoid odontogenic tumors appear as a corticated circumscribed unilocular radiolucency surrounding an impacted tooth (73% cases), most commonly maxillary canines (40% to 60% of cases), lateral incisors, and mandibular premolars [1,2,5]. They may have internal radiopaque foci in two-thirds of cases.⁵ Ameloblastic fibro-odontomas, which are benign mixed odontogenic tumors, present as a well-demarcated unilocular or infrequently multilocular radiolucency containing various amounts of radiopaque material of irregular size and shape [1,2]. Odontomas are tumor of odontogenic origin with proliferation of both epithelial and mesenchymal cells which demonstrate complete differentiation. Radiographically, they may exhibit radiolucency due to a lack of calcification, partial calcification or radiopaque masses surrounded by radiolucent areas [1,2].

When a lesion with an impacted tooth is encountered, clinician should consider these entities in the differential diagnosis. Their radiographic and histopathological findings will help dental practitioners make more accurate diagnoses and create better treatment plans.

This report describes a case of UA of the mandible in an 8-year-old boy that mimics a dentigerous cyst given its unicystic appearance and association with an unerupted permanent mandibular first molar. Surgical enucleation and histopathological examination was performed to establish a definitive diagnosis which revealed plexiform ameloblastoma.

Case Report

An 8 year old boy with chief complaint of swelling on right lower side of face from past 6 months reported to the department of Pedodontics and Preventive Dentistry, PGIDS, Rohtak. The medical history of the patient was non-contributory and no systemic involvement was reported. He was not taking any medication and had no history of any drug allergy. The patient was in mixed dentition stage. On clinical examination, a hard non tender painless swelling extending from mandibular deciduous second molar to retromolar area was noticed. Overlying oral mucosa was normal in texture and appearance. The deciduous molars present on same side were non-carious and any infected periapical pathology was ruled out. All his permanent first molars have erupted except right mandibular first molar on the involved side.

Panoramic radiograph revealed a unilocular well circumscribed radiolucency encircling the crown of unerupted right mandibular first molar with incomplete root formation. The lesion is compressing the developing premolars on the same side and second premolar was somewhat pushed toward the lower border of mandible. CBCT depicted expansion and thinning of buccal cortex in first molar region. Considering the radiographic appearance and association with an unerupted permanent molar, provisional diagnosis of dentigerous cyst, UA and odontogenic keratocyst was made. Blood investigations were done. Under local anesthesia, surgical enucleation of the lesion was done. The unerupted first molar was left undisturbed to provide a chance for self-eruption. The tissue was sent for histo-pathological examination. Histopathology revealed that the tissue section consisted of loosened fibrocellular connective tissue stroma with mild mixed inflammatory cell infiltrate and abundance of engorged vascular channels. On the periphery, interconnecting cords of tumor cells having ameloblast like cells and stellate reticulum like cells in between these layers were present. Diagnosis of Unicystic plexiform Ameloblastoma (intraluminal type) was confirmed. The parents were informed about the condition, treatment options and recurrence potential, after which they provided their informed consent for the conservative treatment and regular follow-up.

The patient was kept on regular follow-up and home hygiene instructions were given. At 3 months follow-up, the bony swelling was comparatively decreased and deciduous second molar was mobile. The patient was totally asymptomatic. At 9 months follow-up, the bony swelling was completely resolved clinically and deciduous second molar was lost. Tip of right mandibular first molar was visible in the oral cavity. CBCT revealed new bone formation and contour of mandibular buccal bone has returned to normal. At 18 months follow-up, complete occlusal surface of permanent right first molar and cusp tip of second premolar were seen. At 36 months follow-up, complete bony healing was seen on CBCT and permanent right first molar and second premolar has erupted without any further intervention. The root formation of right first molar was near completion. The patient was still on regular follow-up and no evidence of tumor recurrence was noticed.

Discussion

Ameloblastoma is the second most common true neoplasm of odontogenic origin, developing from the epithelium involved in the formation of teeth: the enamel organ, epithelial cell rests of Malassez, reduced enamel epithelium, and odontogenic cyst lining [6-8]. Robinson described it as, "unicentric, nonfunctional, intermittent in growth, anatomically benign, and clinically persistent" [6]. Regez and Sciubba reported 11% prevalence of ameloblastoma out of all odontogenic tumors in the jaw [9]. It affects mandible more than maxilla,

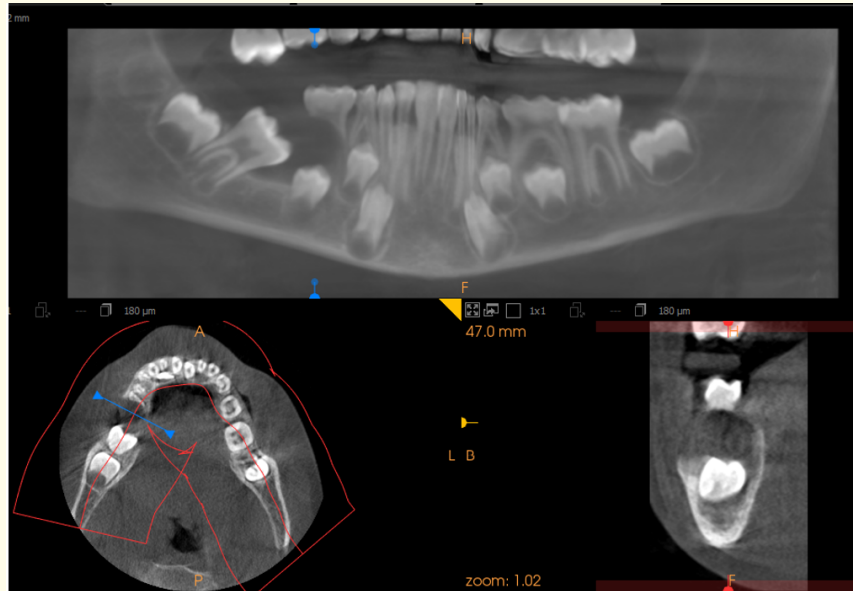


Figure 1: Pre-operative CBCT images showing a unilocular, well-defined radiolucent lesion surrounding the crown of unerupted right mandibular first molar.



Figure 2: Pre-operative CBCT image showing buccal cortical expansion and perforation.



Figure 3: CBCT image showing bone formation and normal contour of buccal and lingual cortical bones at 36 months follow-up.

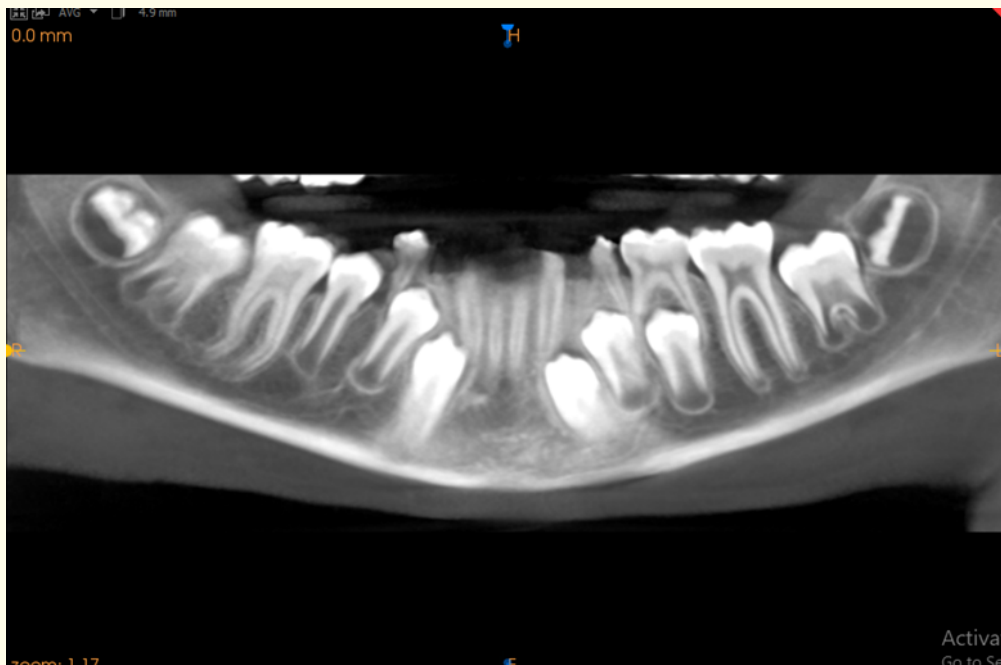


Figure 4: Panoramic view showing new bone formation and erupting right mandibular first molar and premolar at 36 months follow-up.

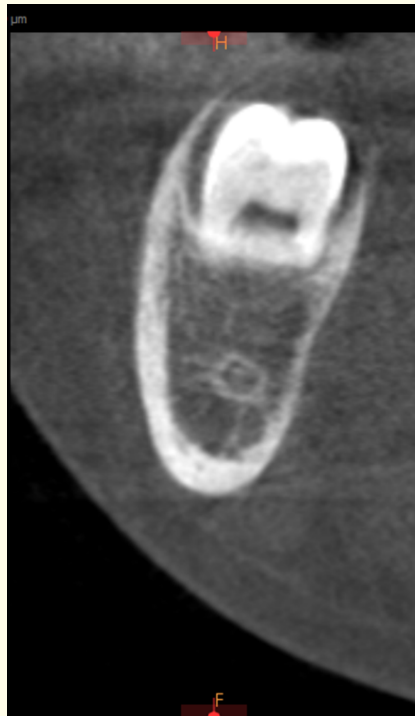


Figure 5: Axial section showing new bone formation and normal contour of buccal and lingual cortical bones at 36 months follow-up.



Figure 6: Clinical picture of the patient showing erupted right mandibular first molar and premolar at 36 months follow-up.

with 77% located in the molar ramus region [10-12]. It is commonly found in adults with its peak incidence in the 3rd and 4th decade of life with a median age of 35 years, and no specific gender predilection (1.14:1.0 male: female) [10]. Occurrence in childhood is very rare.

Clinically, Ameloblastoma is a slow growing, painless expansion of jaw which causes thinning of cortical plates. It can cause displacement and resorption of adjacent teeth, tooth mobility, paraesthesia and dysaesthesia [6,7]. It is considered a benign but locally aggressive polymorphic neoplasm [7].

It can be classified as intraosseous central and extraosseous peripheral types or unicystic and multicystic type [12]. Radiographically, it is seen as a unilocular radiolucent area with a well-defined margin or multilocular with soap bubbles or honeycomb appearance [13]. The multicystic ameloblastoma is the commonest subtype accounting for 86% of all cases compared with unicystic (13% of cases) and peripheral ameloblastomas (1% of cases) [9,10,14].

Unicystic ameloblastoma, is a less aggressive variant of intraosseous ameloblastoma, accounting for 6 - 15% of all intraosseous ameloblastomas [9,10,15]. Robinson and Martinez (1977) first described it as, "cystic lesions that show clinical and radiologic characteristics of an odontogenic cyst but in histologic examination show a typical ameloblastomatous epithelium, lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation" [9]. The prevalence is more in a younger age group [12]. According to a review article on prevalence of ameloblastoma among pediatric population, majority of lesions 91.86% (327 of 356) was found between the age group of 11 and 20 years; only 8.14% (29 of 356) were below the age of 10 years [14]. Another literature review identified 25 out of 513 articles (51 cases in total) presenting cases of UAs in patients under 16 years of age [16].

Ackermann [17] sub-classified UAs as: Group I: Luminal UA (tumor confined to the luminal surface of the cyst), Group II: Intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall) and Group III: Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall and there may be penetration into the surrounding cancellous bone). The luminal and intra luminal UAs can be treated conservatively (enucleation or marsupialization followed by enucleation), whereas UAs showing intramural growths have a high risk for recurrence, require treatment with radical resection (major segmental or en bloc resection with a requirement of 1 - 1.5 cm of clinically and radiographically normal bone and uninvolved margins), as for a solid or multicystic ameloblastoma [9,11,18].

Treatment modalities are dependent on several variables such as size, anatomical location, histologic variant and anatomical involvement [10,14,15]. The treatment modalities can be categorized broadly as: conservative (enucleation, curettage and marsupialization) and radical surgery [14]. According to a study by Lau and Samman (2006) [19] the recurrence rate using different treatment regimen were resection (3.6%), enucleation (30.5%), enucleation followed by Carnoy's solution application (16%) and marsupialization followed by enucleation (18%). Chemical cauterization with Carnoy's solution as proposed by Stoelinga and Bronkhorst, have the ability to penetrate cancellous bone to a depth of 15 mm, and its use following enucleation of UA may result in lower recurrence rates [10].

Majority of cases are mis-diagnosed as dentigerous cysts due to their association with an unerupted tooth. Robinson and Martinez in their study found that 70% of unicystic ameloblastomas were associated with an impacted tooth, and another series showed 83%, although one study showed only 19% related to unerupted teeth [20]. This misdiagnosis can lead to initial treatment with marsupialization. Histopathological examination of the excised tissue can be really helpful to make accurate diagnosis and treatment accordingly.

Several authors have advocated conservative treatment should be first choice in any pathology in children [8]. Radical treatment approach could result in functional and masticatory change, mutilations, and facial deformities. Following factors must be considered

in children: (1) continuing facial growth and different bone physiology (more cancellous bone, increased bone turnover, and reactive periosteum), (2) presence of unerupted teeth, (3) difficulty in the initial diagnosis, and (4) psychological development of child [8,10,20]. Therefore, conservative treatment should be considered in children to provide reasonable cosmetic and functional outcome which also offers better quality of life, but the recurrence rate with this treatment may be higher [10]. Therefore, the patient should be kept on longer follow-up to assess any recurrence or any untoward changes because > 50% of recurrences occurs within 5 years of the treatment [8,12]. Some authors consider recurrence is not an important consideration and should not be considered as equivalent to failure because a second surgery can be successful [18]. We treated our patient by conservative management and no complications were reported, without any signs of recurrence.

The involved teeth are usually extracted together with the tumor during surgery, to prevent their recurrence [8]. However, tooth loss can result in psychological, functional and esthetic problems in young children. Thus, the impacted tooth within ameloblastomas was preserved and spontaneous eruption occurred and is functioning well.

Conclusion

Different pathologies involving an unerupted tooth must be considered during patient evaluation and histopathology and radiographic examination play an important role in definitive diagnosis. The management of UAs should be planned taking in consideration the age, physical growth and psychological development of child.

Conflicts of Interest

Nil.

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Nil.

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