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A case of dentigerous cyst in a pediatric patient - With an insight into differential diagnostic entities

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ABSTRACT

Dentigerous cysts are one of the most common developmental odontogenic cysts involving the unerupted or impacted tooth. Most frequently seen in 20–30 years of life. Cases which have been reported within 10 years of life, in mixed dentition period are hardly few in number. Here we present an interesting case of dentigerous cyst in a pediatric patient involving unerupted permanent mandibular right second premolar with an insight into differential diagnostic entities of dentigerous cyst. Complete removal of the cyst along with attached tooth structure was done under general anesthesia. Careful evaluation of the patient with past medical history, clinical, radiographic and histopathological examination would help the clinician in early diagnosis to administer appropriate treatment.

1. Introduction

A dentigerous cyst (DC) is one that is formed by follicle expansion of an unerupted tooth enclosing its crown [1]. Accounting for 25% of all the cysts, making it one of the frequently occurring developmental cysts of jaw [2]. Although noticed in wide age range, most commonly seen in between 20 and 30 years of life, less frequently below 10 years of age [3]. Majority of the cases shows association with the impacted or unerupted mandibular molars, second most common site is maxillary canines followed by maxillary molars [4]. Most of the DCs are painless unless secondarily infected, however mainly noticed during routine radiographic examination [5]. This case report presents a rare case of DC in a 10 year old girl with mixed dentition involving unerupted right mandibular permanent second premolar. Larger DCs sometimes can resemble aggressive lesions like keratocystic odontogenic tumor and ameloblastoma. Entities like unicystic ameloblastoma (50% of the cases) and ameloblastic fibroma (75% of the cases) have similarities in some aspects to DC such as, predilection to occur in children and young adolescents and frequently seen in association with unerupted or impacted tooth (75% of ameloblastic fibromas) [6–8]. A careful evaluation of clinical, radiological and histopathological differential diagnosis is needed before scheduling the surgery. The timely diagnosis and treatment should be done as untreated cases of dentigerous cyst can lead to complication such as, bone deformation, loss of permanent tooth and may also develop into odontogenic tumors and carcinomas [9].

2. Description of the case

A 10 year old girl reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of a asymptomatic swelling and difficulty in mastication on the right side of mandibular region since 3 months. On inspection, the patient was healthy. Medical history was not relevant. Intraorally there was a bony swelling present opposite to right primary first molar occluding the buccal vestibule measuring of about 3 × 3 cm. There was no caries or periodontal problem.

3. Investigations and differential diagnosis

Clinical differential diagnosis included periapical cyst, keratocystic odontogenic cyst (KOT) and ameloblastoma. As there was no carious lesion seen clinically, periapical cyst was ruled out.

Orthopantamograph (OPG) was advised. The OPG findings showed, a well-defined unilocular radiolucent area in association with the crown of an unerupted second permanent premolar with diffuse corticated border. On the mesial side of the unerupted tooth the lesion was slightly seen below the cementoenamel junction (CEJ). Root resorption was seen

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with respect to right primary second molar and permanent first premolar (Fig. 1).

The most favourable radiographic differential diagnosis for this case included DC, ameloblastic fibroma, unicystic ameloblastoma, KOT, and adenomatoid odontogenic tumor (AOT).

DC was the first choice of diagnosis as the radiograph revealed unilocular radiolucency surrounding the neck of the crown of an unerupted tooth, with diffuse and thin corticated borders, which are the radiographical features usually seen in DC [9–11]. DC has to be differentiated from the hyperplastic follicle. If the follicular space is above 5 mm DC can suspected, as the normal follicular space is 2–3 mm [9].

But other entities like ameloblasctic fibroma, unicystic ameloblastoma, KOT and AOT has to be ruled out before considering the treatment options.

Ameloblastic fibroma is one of the differential diagnosis for this case, as OPG showed unilocular radiolucency surrounding the neck of the tooth which was slightly below the CEJ on the mesial side. Usually ameloblastic fibroma appears as a unilocular radiolucency and sometimes multilocular. It usually shows well demarcated sclerotic border and sometime associated with unerupted or displaced tooth [12].We had to wait for the histopathological findings to rule out ameloblastic fibroma.

Unicystic ameloblastoma (UA) of dentigerous variant shows unilocular radiolucency in association with an impacted tooth. The involved tooth mainly would be mandibular third molar followed by mandibular canines in case of unicystic ameloblastoma. Knife edge root resorption and destruction of the anterior border of the ramus are also the characteristic features of the UA [13,14].Unicystic ameloblastoma was also one of the probable and important radiographical differential diagnosis for this case. To rule out this entity histopathology report was needed.

KOT radiographically seen as unilocular or multilocular radiolucency surrounded by corticated margins with slight tendency towards unilocular radiolucency. Where 30% of the cases are associated with the unerupted teeth, mainly third molars. The DC of bigger size always pose problem in diagnosis between KOT and DC. One peculiar feature of KOT is antero-posterior expansion with considerable mesiodistal extension [15]. As our case showed the unilocular radiolucent area surrounding the crown of an unerupted permanent second premolar with mesio distal expansion, KOT was ruled out.

Radiographic findings of AOT sometimes resemble dentigerous cyst. Follicular type of AOT which is seen in association with the unerupted tooth could be the potential differential diagnostic entity in this case. AOT is seen as unilocular radiolucency or sometimes mixed radiopaque - radiolucent lesion with well defined sclerotic or corticated border. The unerupted tooth is canine in 70% of the cases, permanent premolars are rarely involved [16]. In our case unerupted premolar was involved and the lesion showed diffuse corticated border involving the neck of the unerupted second permanent premolar. So, AOT was ruled out but the diagnosis has to be confirmed by histopathological features.

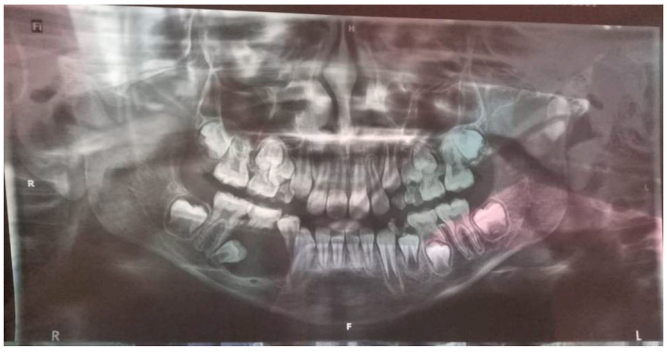

Fig. 1. Orthopantomogram showing unilocular radiolucent lesion associated with erupting permanent second premolar.

In correlation with the clinical and radiographic features interim diagnosis of dentigerous cyst or ameloblastic fibroma or unicystic ameloblastoma was made. However, the diagnosis has to be confirmed by histopathological findings.

Considering the interim diagnosis the preferred treatment option was the enucleation of the cyst. Before the surgery, blood and urine examinations were recommended, and the values were normal. The surgical resection was done under general anesthesia. Complete removal of the cyst, along with the tooth attached to it was done (Fig. 2). Gross specimen which consisted of lining of the lesion along with the attached tooth i. e right permanent mandibular second premolar was sent for histopathological examination. Cystic cavity did not show any vascular content or a cheesy material or clear fluid. The cystic cavity was packed with sterile Abgel gauze to attain hemostasis and to inhibit hematoma formation followed by suturing (4-0 vicryl resorbable suture).

Histopathology report showed the presence of cystic lumen lined by epithelium lined by a 2 to 3 layers of non keratinized stratified squamous epithelium covered by connective tissue stroma showing scattered inflammatory infiltration (Fig. 3). DC occasionally shows odontogenic rests in the connective tissue, then histopathological differential diagnosis would include lesions like ameloblastic fibroma, odontogenic myxoma or an unicystic ameloblastoma [9]. As there were no odontogenic components seen in our case, these entities were ruled out and diagnosis of dentigerous cyst was given on correlation with clinical, radiographic and histopathological features.

4. Discussion

DCs are the most prevailing developemental odontogenic cysts. Pathogenesis of DC may be either developmental or inflammatory. In case of developmental origin there is a collection of fluid between the tooth enamel and reduced enamel epithelium. In inflammatory origin, periapical infection from the overlying necrotic primary tooth spreads to contain the sac of the unerupted permanent successor [17].

Accurate diagnosis of DC is of extreme importance and lesion has to be carefully evaluated clinically, radiographically and histologically to rule out locally aggressive entities like KOT, ameloblastoma, ameloblastic fibroma and odontogenic myxomas before concluding the diagnosis.

Differentiating features of the lesions are given in Table 1.

Review of literature showed that age of occurrence of DC is in the 20–30 years of life and with male predilection [18]. Reports of its occurrence within 10 years of life are very few [1]. In our case the patient was 10 year old female, which makes this case unique as occurrence of DC in the first decade, in mixed dentition period is less common.

The most common site of occurrence of the cyst is mandibular region. Rarely anterior region of maxilla has also been reported [1,19]. In

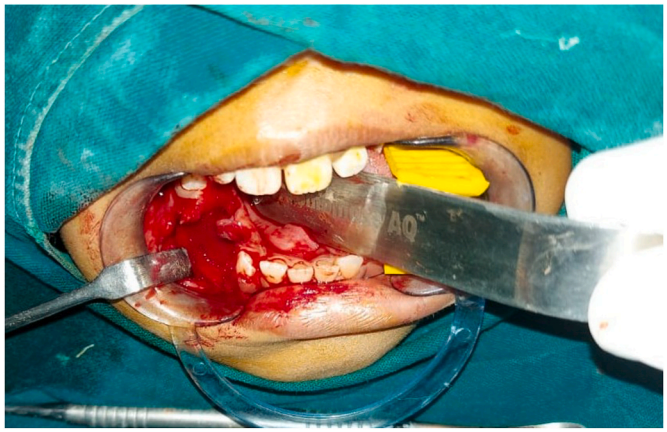

Fig. 2. Intra oral image showing cystic cavity.

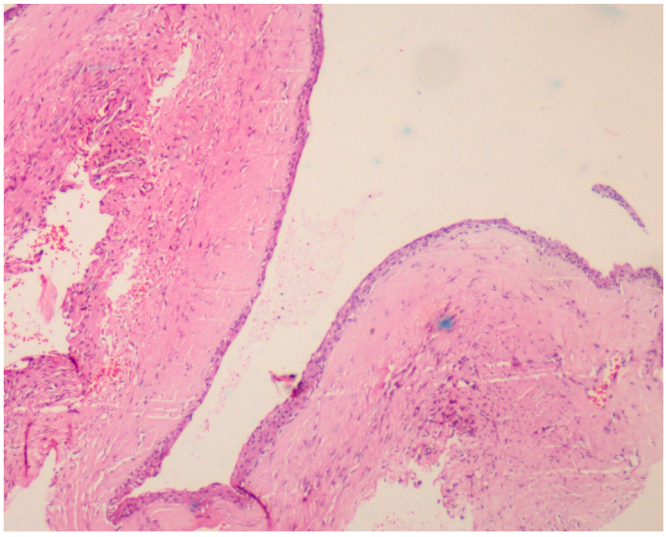

Fig. 3. Hematoxylin and Eosin stained image showing 2–3 layer of non-keratinized stratified squamous epithelium with connective tissue stroma.

present case the site of the lesion was right mandibular region, which was consistent with the review of literature.

Most of the DC are unilateral and singular. Association of syndromes like basal cell nevus syndrome, Mucopolysaccharidosis, Cleidocranial dysplasia can be expected when they are present in multiple number and present bilaterally [9,18]. Our case was non syndromic and there was presence of unilateral solitary lesion on the right side.

DC usually presents as a asymptomatic swelling involving bone expansion if secondarily not infected [9,20]. However, they can enlarge to a considerable size leading to severe root resorption, tooth displacement, facial asymmetry, hollowing out of the bony structure [9]. Here the patient gives history of slowly enlarging swelling over a period of 3 months without any symptoms.

Radiologically DC represents as unilocular radiolucenct lesion sometimes multilocular. Three radiographic variants of the cyst are central varient, circumferential type and the lateral type [9,14].Our case showed central variant where radiolucency was seen covering the neck of the unerupted tooth. Other variants like circumferential variant is seen as a cyst enveloping the entire tooth, and lateral variant is seen as a radiolucency seen on one aspect of the teeth [9]. Resorption of root of right primary second molar and permanent right first premolar was noticed.

Microscopically DC shows a cystic lumen lined by 4–5 layer of non keratinized stratified squamous epithelium. Retepeg formation with increased thickness of epithelium is seen in case of inflammatory cyst. The underlying connective tissue capsule will be collagenized or myxomatous and foci of inflammatory cells may be seen in case of 7 inflammation. Islands of odontogenic rests can also be seen in the connective tissue [6,19]. Epithelium of DC has the capability to go through metaplastic changes if left untreated [9].

Histopathology report of our case showed cystic lining of 2–3 layer of nonkeratinized stratified squamous epithelium surrounding cystic lumen. Stroma showed sparsely arranged collagen fibers and mild inflammatory infiltration was seen.

Treatment modalities for DC depends on the patient's age, situation of the cyst, tooth locality, and root formation. Most commonly used treatment options are complete enucleation and marsupilization with removal of the associated tooth [6]. In our case we chose enucleation of the cyst as a treatment option. Cystic lining was completely removed. Involved unerupted second permanent premolar was removed along with primary second molar, in order to prevent recurrence. Cystic cavity was filled with abgel gauze to attain hemostasis and to inhibit formation

Table 1
Differentiating features of lesions [1,9,10,12,13,15,16,19].

FEATURES	DENTIGEROUS CYST	RADICULAR CYST	KOT	UNICYSTIC AMELOBLASTOMA	AOT	AMELOBLASTIC FIBROMA
AGE	2nd to 3rd years of life	2nd–5th decade	2nd–4th decade	2nd decade of life	Within 30 years of age	First two decades of life
SEX	Male predilection	Not specific	Male	Male predilection	Female predilection	Female predilection
SITE	Posterior mandible	Apex of any non vital tooth.	Posterior mandible	Posterior mandible	Anterior maxilla	Posterior mandible
ASSOCIATION WITH UNERUPTED TOOTH	Yes	No	Some cases	Yes	Yes	Yes
RADIOGRAPHIC FEATURES	Unilocular radiolucency Showing 3 variants Central Circumferential Lateral variant	Round or ovoid radiolucent area	Usually unilocular may be multilocular with well defined or scalloped border and anteroposterior expansion	Unilocular radiolucency with well circumscribed borders with associated unerupted or impacted tooth.	Intra osseous AOT shows well circumscribed unilocular radiolucency or mixed appearance (radiopaque-radiolucent) with well circumscribed corticated or sclerotic borders. And associated with the impacted or unerupted tooth.	Smaller ones show uniclocular radiolucency, larger ones are multilocular. With well defined and sclerotic border.
HISTOLOGIC FEATURES	Cystic lumen lined by 4–5 layer of non keratinized stratified squamous epithelium. The capsule may be fibroid or myxoid with foci of inflammatory cells may be seen in case of inflammation.	Cavity lined by stratified squamous epithelium and covered by a connective tissue stroma containing a predominantly chronic inflammatory infiltrate and cholesterol clefts.	Stratified squamous epithelium of 6–8 layer thick with basal cell layer showing picket fence appearance.	Ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth	Epithelial cells arranged in 1.spindle shape, in the form of whorls, nests, and bundles, 2.cuboidal shape arranged in duct like structures. Supported by a thick fibrous connective tissue capsule.	Odontogenic epithelium arranged in cords, nests and small islands in scanty cytoplasm, with stallate reticulum like cells in the center.

of hematoma and sutures were placed. Prosthesis placement has been planned for the missing tooth.

5. Conclusion

DCs are very uncommon in pediatric patients and, in the first decade of life with mixed dentition, an undiagnosed and untreated dentigerous cyst can lead to potential complications. A better prognosis can be expected in children as they have greater potential to regenerate bony structure than adults therefore, a thorough and timely evaluation of the patient history coupled with clinical and radiographic examination would help in early diagnosis and treatment.

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Declaration of competing interest

None.

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