

Case Report

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Sequalae of Dentigerous Cysts on the Dental Development of Permanent Teeth in Children: A Case Report

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ABSTRACT

The dentigerous cyst is considered as a benign lesion. However, as growing, it can damage the anatomical structures surrounding it and influence the developing neighboring dentition which was the observed in our case where the radicular formation of the neighboring permanent tooth was arrested. This was the consequence of the delayed diagnosis of the cystical lesion which had silently taken a huge size before getting discovered through a routine radiographic examination.

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Introduction

Also known as a follicular cyst, a dentigerous cyst is an odontogenic cyst resulting from fluid accumulation between reduced enamel epithelium and the crown of an unerupted or developing tooth [1]. Dentigerous cysts (DCs) are considered as the most common developmental cyst of the jaws and the second most commonly encountered odontogenic cyst after radicular cysts [2]. They are mostly sent at the age of 20-30 years and less commonly observed in children with a prevalence of 4-9 % occurring under the age of 10 years, rarely in elderly patients [3,4]. These entities have a slight male predilection and the mandibular third molar is the most involved tooth followed by the maxillary third molar, maxillary canine and mandibular second premolar [5,6]. They are rarely observed in incisors and supernumerary teeth [2]. Their exact etiology is still unknown and most of them are considered developmental [6]. Some cases of DCs have of an inflammatory origin resulting of the propagation of the inflammatory process from the overlying primary tooth [7].

Gener Ally not accompanied with clinical signs, these entities grow silently and are not discovered until they get a considerable size manifesting in the appearance of a jugal swelling, facial

asymmetry, paresthesia of the inferior alveolar nerve or pain once they get infected. Also, they can be accidentally detected during a routine radiographic examination. Thus, their diagnosis is often delayed compromising consequently their prognosis. The radiographic appearance of such a pathology is pathognomonic presented as a radiolucent typically unilocular, rarely multilocular Shaowu surrounded by well-defined margins, unless in case of infection, the radiographic borders of the lesion appear to be ill-defined. When the DC shows a multilocular feature, the differential diagnosis must be made so as to exclude other lesions that can be associated with an impacted tooth and might have the same radiographic appearance such as ameloblastoma, odontogenic keratocyst, and odontogenic adenomatoid tumor justifying the ultimate née of the histopathological examination since the radiographic examination is not sufficient to discriminate between these lesions [8,5].

When developing, DCs can damage the neighboring structures. This paper aims to illustrate the sequelae of DCs on the developing dentition.

Case History

A 7-year-old girl in good health condition with no medical antecedents nor hospitalization was referred to the Pediatric and Preventive Dentistry Department by the Oral Medicine and

Surgery Department to which she was referred by her dentist. The Panoramic radiography (Figure 1) showed the presence of a well-defined bilocular radiolucent lesion surrounding the crown of the mandibular right second premolar (45) which appeared to be floating within the lesion which was huge. The radiological appearance of the tooth seemed to be different compared to the other premolars. Also, the panoramic radiography showed the root resorption of the mandibular right first primary molar (84) and the encompassment of the mesial root of the mandibular right permanent first molar (46) in the lesion. The patient was also subjected to a Cone Beam Computed Tomography (CBCT) examination in order to define the dimensional extension of the lesion and it showed that there was a rupture of the external bony cortical (Figure 2a&b).

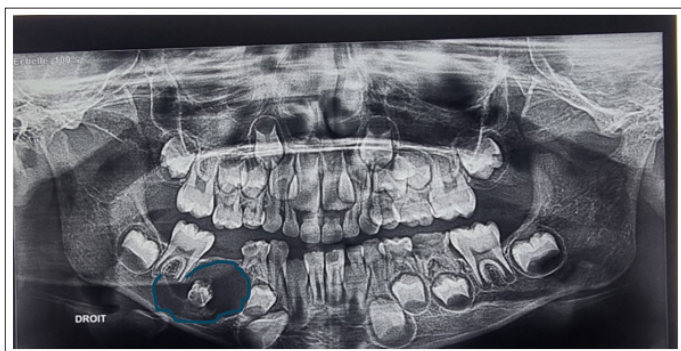


Figure 1: Pre-Operative Panoramic X ray Demonstrating the Presence of a huge Radiolucent Multilocular lesion with Well-Defined Margins, Encompassing the crown of the 45

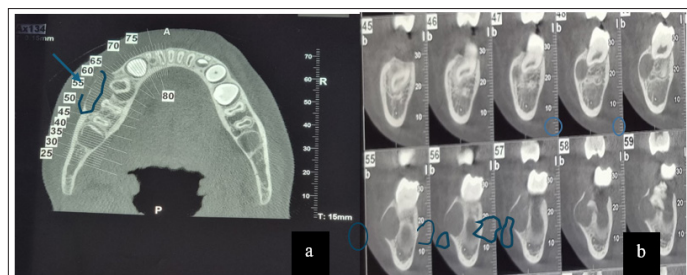


Figure 2 : (a&b) : CBCT Scan showing the rupture of the External Bony Cortical as a Result to the Extension of the Cystic Lesion

Differential Diagnosis, Interventions and Treatment

Although the lesion was suspected to be a dentigerous cyst, the differential diagnosis must be made so as to eliminate the diagnosis of some lesions that have the same radiographic appearance such as hyperplastic dental follicle, radicular cyst of the primary tooth surrounding the crown of the underlying permanent tooth, odontogenic keratocyst, unicystic ameloblastoma, ameloblastic fibroma, odontogenic adenomatoid tumor and calcified odontogenic cyst [8-10]. Thus, the marsupialization technique was applied in association with the extraction of the 84 by the Oral Medicine and Surgery Department team and the histopathological examination of the specimen confirmed the diagnosis of a DC in inflammatory remodeling (Figure 3). A removable prosthesis was confected so as to replace the missing teeth (Figure 4). A palatal expander was performed in order to expand the maxillary arch (Figure 5) and the patient was maintained under regular follow-ups.

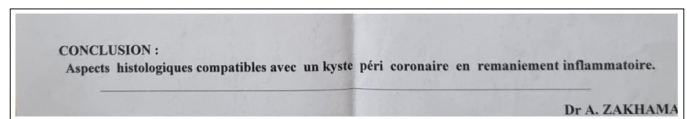


Figure 3: The Histopathological Examination Confirmed the Diagnosis of a Dentigerous Cyst in Inflammatory Remodeling.



Figure 4: A Removable Prosthesis Replacing the Missing Teeth was Confected

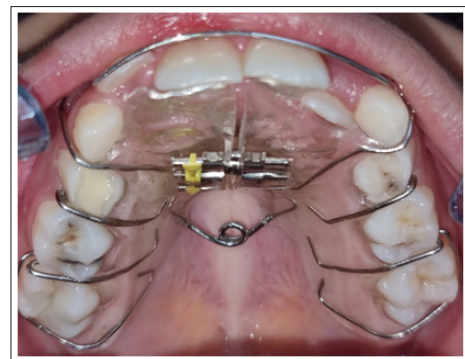


Figure 5: A palatal expander was done in order achieve the transverse expansion of the maxillary arch

Conclusion and Results

One year later, the clinical monitoring of the patient revealed the beginning of eruption of the 45 which appeared blackish (Figure 6 a, b&c). During the next follow-up appointment, there was a swelling located at the mandibular right region associated with pain revealing the recurrence of the cystic lesion. A complete enucleation of the lesion associated with the extraction of the tooth were done and the patient was kept under a continuous monitoring (Figure 7). A Two-year-follow-up showed a complete healing of the lesion with bone regeneration and a normal development of the 44. Also, it revealed the arrest of root development of the mesial root of the 46 (Figure 8). The carious lesion of the 46 was treated so as to preserve the pulp vitality of the tooth and avoid the root canal treatment. Moreover, the treatment of the decayed maxillary primary molars is being processed and the patient is still under regular monitoring for both, the upper expansion appliance and the lower removable prosthesis. An implant rehabilitation procedure is planned to be applied at adulthood so as to replace the 45.

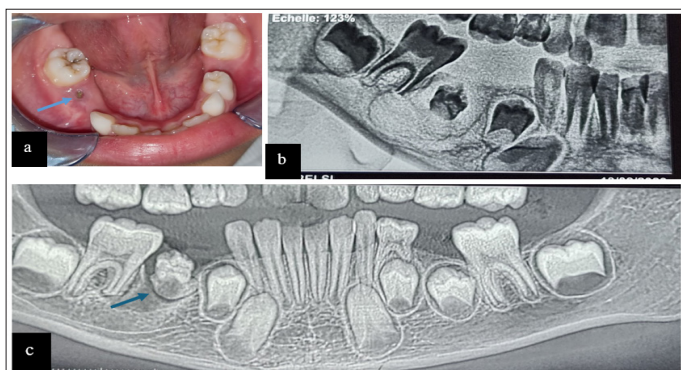


Figure 6(a): The Clinical follow-up of the Patient revealed the Eruption of the 45 which appeared Blacklist

Figure 6(b&c): The Radiographic Follow-up Showed the Eruption of the Tooth without Root Formation



Figure 7 : A Complete Enucluation of the Lesion Associated with the Extraction of the Tooth were done

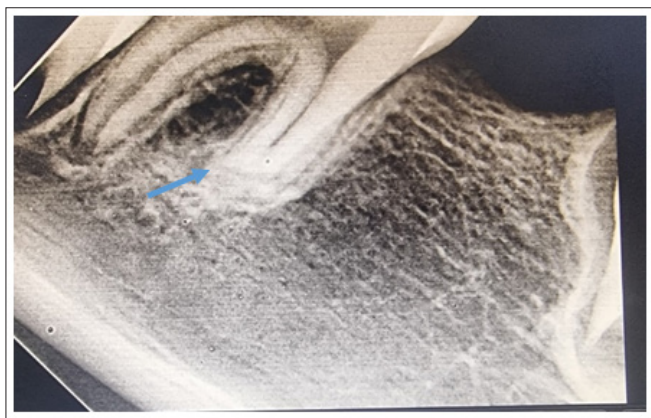


Figure 8: The Periapical X-Ray Demonstrated a complete Healing of the Lesion with Bone Regeneration. It Showed also the Arrest of Root development of the Mesial Root of the 46

To sum up, DCs are considered as benign pathologies that can be encountered at the first decade. Being usually asymptomatic, these bone lesions grow in silence and can acquires a huge size before getting discovered, which makes their diagnosis often delayed. Although their identification is often incidental, through a routine radiographic examination, they can be detected once they become symptomatic manifesting in the appearance of swelling, pain, paresthesia of the inferior alveolar nerve and facial asymmetry. The therapeutic modalities of DCs depend on various factors including the extension of the cyst and must take into account the neighboring anatomical structures. The histopathological examination is primordial so as to confirm the prior diagnosis and

exclude more aggressive lesions that can have similar clinical and radiographic signs. The possible recurrence and the neoplastic potential of such a pathology justify the necessity of regular follow-up. The disturbance of the developmental and eruptional potential of the nearby dentition is a possible sequela of DCs. Therefore, the earlier the diagnosis of the cystic lesion is, the lower its extension will be and the more their sequelae will be avoided.

Discussion

DCs are a slow growing, benign cysts that arise from the dental follicle of an unerupted tooth, enclosing its crown and attaching to it at the level of the cemento-enamel junction. The majority of DCs are solitary. However, they can be bilateral or multiple which are found associated with syndromes such as cleidocranial dysplasia and Maroteaux-Lamy syndrome. A rare and unique case of DCs involving all the four quadrants and not associated with any syndrome, was reported by Aher V et al. 2013, in a 24-year-old male patient. In such a case, the DCs were associated with third maxillary and mandibular molars with the presence of supernumerary teeth in the mandibular cystic lesions [11].

From a histopathological viewpoint, DCs are characterized by the presence of a loose fibrous tissue lined by a thin regular nonkeratinized stratified squamous epithelium developed of remnants of reduced enamel epithelium around the crown of an unerupted or impacted tooth, which undergo cystic degeneration with fluid accumulation leading consequently to the formation of the cyst. Once it becomes inflammatory, the DC contains an inflammatory cellular infiltrate along with hyperplastic epithelium [12].

Clinically, small cysts are generally asymptomatic and painless. Thus, their identification can be either incidental during a routine radiographic examination, or occasional from delay in the eruption of a permanent tooth [13]. DCs can grow mysteriously until they acquire a remarkable size or get secondarily infected, usually as a result of the spread of the periapical inflammation from an overlying primary tooth. At that moment they become symptomatic being associated with swelling, mild sensitivity, tooth mobility, and tooth displacement once it acquires a size higher than 2 cm in diameter resulting in a palpable mass. DCs can enlarge leading to causing pathologic mandibular fracture and facial asymmetry. However, the paresthesia of the inferior alveolar nerve is their most commonly encountered symptom [2]. Radiologically, DCs are typically presented as a radiolucent lesion along with a well-demarcated sclerotic border surrounding the crown and extending along the root surface of an unerupted or impacted tooth which appears entirely enclosed within the cyst. They typically present a unilocular radiological appearance but, they can show a multilocular aspect when they develop in areas of different bone densities [2,3,8]. The radiological resemblance of DCs with many lesions including hyperplastic dental follicle, radicular cyst of the primary tooth surrounding the crown of the underlying permanent tooth, odontogenic keratocyst, unicystic ameloblastoma, ameloblastic fibroma, odontogenic adenomatoid tumor and calcified odontogenic cyst may confuse dental practitioners but here are some specific characteristics that can better lead to the diagnosis of DCs : attachment to the cemento-enamel junction, the infiltration of the neighboring tooth, bubble-like cortex, and the pericoronal space thicker than 5mm. In comparison to DC, the odontogenic keratocyst has a slightly denser radiological content. On the other hand, the differential diagnosis between DC and unicystic ameloblastoma or ameloblastic fibroma cannot be established radiologically because of the absence of an internal structure [9].

In the present case, the pathological lesion was discovered through the presence of a palpable mass developed in the buccal surface of the mandibular right quadrant next to the placement of the lower right second premolar. This region is particularly characterized by the close distance between the mandibular second primary molar and the follicle of the successor permanent tooth which can explain the rapid propagation of an inflammatory process from the apical region of the necrotic primary tooth to the follicle of the developing premolar leading to the arising of an inflammatory originated DC [14].

This probably was the case in our patient for which the histopathological examination confirmed the presence of an inflammatory infiltrate in the specimen. However, the mandibular right primary second molar was extracted at the first visit. That's why we cannot certainly consider that her cystic lesion was basically inflammatory or secondary infected.

Radiologically, the lesion was bilocular encompassing the crown of the 45 which appeared floating within the lesion, corresponding to the circumferential variety of DCs described by Shear and Speigh. In fact, they described three radiological types of DCs: the central variety characterized by the DC envelops symmetrically the crown, the lateral variety where the tooth is surrounded by the cyst on only one side, and the circumferential variety in which the whole tooth seems to be englobed by the cyst [2]. Although they are non-neoplastic, DCs are known to have a neoplastic potential in which way their epithelial lining may develop benign tumors such as ameloblastoma, adenomatoid odontogenic tumor, or complex odontoma or malignant tumors such as mucoepidermoid carcinoma and squamous cell carcinoma [2]. Also, DCs can coexist, distinctly, with other odontogenic lesions at the very same radiographic area such as the Calcifying Epithelial Odontogenic Tumor (CEOT), the case that was reported by Kurup RJ et al. 2022 where the final diagnosis retrieved of the clinical, radiographic and histological examination, supposed the simultaneous existence of a DC and a CEOT along with an impacted permanent right mandibular first molar in a 17-year-old patient [15].

The therapeutic management of DCs takes into account different criteria that must be well evaluated before choosing the most appropriate treatment, such as the size and location of the cyst, the removal of unerupted tooth, the follow-up possibilities, the degree of bone destruction that can be caused by the surgical technique, and the patient characteristics [16].

Tovio-Martínez E et al. 2022 proposed a therapeutic algorithm for the management of DC based on their initial size. According to them, lesions under 1 cm of diameter must be treated by curettage, those whose diameter is between 1-7 cm their treatment must combine enucleation and curettage. For lesions with a 7-8 cm diameter, marsupialization with or without adjuvant treatments is the best therapeutic option. For all the previous lesions, the treatment can include or not the extraction of the involved tooth. However, cystic lesions having a diameter that reaches or overtakes 11 cm, must be managed by adjuvant resection and treatment with extraction of the etiological tooth [17].

Whenever possible, surgical enucleation of the cyst along with histopathological examination is considered the treatment of choice while considering the neoplastic transformation potential of DCs. However, this technique is considered mutilating since it is associated with a huge bone destruction. Thus, other minimally invasive treatment options including decompression and marsupialization, have been suggested so as to protect as possible,

the surrounding anatomical structures together with preserving the causal tooth [7]. Decompression and marsupialization are advocated for extensive cysts when the full elimination of the encompassing structure is mutilating for a cyst of considerable size. Also, they are especially used as conservative approaches in situations where an accurate pre-operative diagnosis has been established using FNAC (Fine Needle Aspiration Cytology) which is a fast, non-invasive, safe, and easy to perform surgical method consisting on aspirating the cysts contain using a needle inserted into the lesion through the resorbed thinned vestibular plate, followed by performing the histopathological examination of the aspirate. This technique is considered as efficient allowing to get closer to the histopathological diagnosis prior to surgery making possible to apply from the beginning, the most conservative surgical approach [16]. The surgical management of DC must minimize as possible the post-operative complications. This strongly depends on the time taken for the wound closure after the surgical intervention. In order to fasten the healing of tissues and protect the operated areas, silk sutures have been used all over the years. A recent study has proved that cyanoacrylate, as tissue adhesive used for wound closure by creating a barrier that prohibits the infection of the operational zone, is more effective in reducing erythema and facial swelling and enhancing health related quality of life of patients compared to traditional silk suture. However, this newly invented technique was found to induce more post-operative pain compared to the traditional one [18]. The present case was initially treated by marsupialization as a therapeutic approach aiming to preserving the involved tooth (the 45) and avoid bone mutilation taking into account the huge size of the cyst. This technique particularly requires a close follow-up in order to detect any complications. Indeed, one year later, the 45 started its eruption through the mucosa and appeared strangely blackish. The panoramic radiography showed the eruption of the tooth without root formation. Soon after the manifestation of the tooth in the oral cavity, a painful swelling appeared at the same ancient location of the cyst revealing its recurrence. Thus, the complete enucleation of the lesion and the extraction of the involved tooth were performed. Traditional silk sutures were used so as to protect the enucleation area and facilitate the recovery of the cystic lesion. A removable prosthesis was confected in order to replace the missing teeth including the 85, the 84 and the 75, which the patient continued to wear pending the implant rehabilitation at adulthood. The prosthesis will be redone as long as the dental adjacent is proceeding.

In dentigerous cystic pathology, the healing follow-up is primordial so as to supervise the bone ossification and the eruption of the causal tooth. Moreover, the potential of recurrence and neoplastic transformation of such a pathology makes the monitoring ultimately necessary [16].

A case of transformation of a DC into a squamous cell carcinoma was described by Zapala-Pospiech A et al. 2013 in a 66-year-old male patient. They reported that the malignant transformation of a DC's epithelium might be caused by its chronic inflammation [19].

DCs can leave prejudicial sequelae mentioning essentially the extraction of the involved dentition, the displacement of the adjacent structures such as the inferior alveolar canal and the displacement or root resorption of the developed neighboring teeth which was found in cases reported by Perez A et al. 2022 [9].

The developing dentition can also be damaged by the development of such lesions which was the case of our patient. In fact, the periapical radiography done two years after the discovery of the

cyst revealed the arrest of root development of the mesial root of the 46 which appeared shorter compared to the distal one. Therefore, the carious lesion of the 46 was treated as soon as it appeared so as to preserve the pulp vitality of the tooth and avoid the root canal treatment. Fortunately, the development of the 44 and its eruption are normally proceeding and there was an optimal healing of the lesion with a complete bone regeneration. The present case highlights the ultimate importance of follow-up of the development of the adjacent teeth and their eruption that can be damaged by such a dentigerous cystic lesion.

Ethical Considerations: A written informed consent was obtained from the patient's parents.

Conflict of Interest: No conflict of interest was declared by the authors.

Authors Contributions:

Faten Balloumi: Writing (Original Draft), Writing (Review and Editing).

Marwa Chatti: Supervision.

Abir Mannai: Visualization.

Ahlem Baaziz: Supervision.

Data Availability Statement

The data that support the findings of this paper are available from the corresponding author, upon reasonable request.

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