Management of permanent teeth in dentigerous cysts in children: a case report

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Abstract

Dentigerous cysts (DC) are the second most common odontogenic lesion, after radicular cyst, with a prevalence ranging from 0.8% to 3.6% in general population. DC mainly affect people in second and third decades of life, with a male predominance. Because diagnosis is often late, enucleation and avulsion of the impacted tooth are often necessary.

However, when a dentigerous cyst is discovered early in a child with delayed tooth eruption, the aim of the treatment is to preserve and position the permanent tooth on arches. In such cases, conservative therapies such as cystic decompression may be appropriate.

We report a case of a DC in a 10-year-old child with delayed eruption of teeth 22 and 23, managed by decompression alone and ortho-surgical traction allowing the impacted teeth to be placed on the arch. This article highlights the importance of multidisciplinary surgical and orthodontic management of dentigerous cysts in children, and patient’s compliance to treatment.

Categories: Pathology, Dentistry, Oral Medicine

Keywords: mixed dentition, orthodontic treatment, cyst decompression, conservative treatment, unerupted teeth, dentigerous cyst, case report

Introduction

Dentigerous cysts (DC) are part of inflammatory odontogenic cysts due to an included or impacted tooth according to the 2017 OMS classification [1] and represent 24% of all epithelium lined jaw cysts, with a male predominance. Peak incidence is in second and third decades of life [2]. DC affect in a decreasing way wisdom tooth, maxillary canine region and second mandibular bicuspid. Usually discovered incidentally, DC are benign tumors, encompass the crown of unerupted tooth and can grow a very large size.

Various therapeutics exist and are used with a therapeutic gradient according to patient’s age, localization of the cyst and the tooth implicated. Conservative techniques such as marsupialization or cystic decompression are indicated particularly in children with immature permanent teeth contained within DC [3]. Marsupialization was first described by Partsch [4] who performed a large window kept open by suturing the cystic membrane to surrounding soft tissues of oral cavity, so as to promote cystic drainage. Otherwise, decompression takes up the principles initiated by Partsch but drainage is carried out by a smaller mucous window held open by an adjoining device, communicating the cystic contents and the oral cavity [5]. Often confused by their nature, the objective of these techniques is the same: to keep the cystic contents open so as to promote constant drainage involving a progressive decrease in intracystic pressure and thus promote its regression and bone regeneration. When permanent teeth in children are included and repressed, the challenge itself is no longer only bone regeneration but the hope that tooth will be able to function on the arch through favourable tooth movement and root formation.

This case has been written in accordance with CARE (2017) guidelines for cases reports [6]. The aim of our case focused on a 10-year-old patient presenting with a dentigerous cyst located in the anterior maxillary region, affecting the eruption of two permanent teeth. In this context, we opted for a conservative treatment approach. The complex challenge in this case was the management of this dentigerous cyst and correct positioning of these two permanent teeth on the dental arch to ensure both aesthetics and function.

Case Presentation

A 10-year-old asymptomatic patient with no particular medical or surgical history, consulted the oral medicine department of the Reims University Hospital for an orthodontic consultation due to unilateral uneruption of the lateral incisor.
Extra-oral clinical examination revealed no abnormalities (no swelling, no lymphadenopathy). Intra-oral clinical examination (Figure 1) revealed the persistence of teeth 62, 63 and 64 with a distoversion of 21 and a slight vestibular swelling next to the periapical area of teeth 62 and 63, which was painless to palpate. Temporary teeth 62, 63 and 64 do not show any infectious signs (positive cold pulp test and physiological percussion test).

**FIGURE 1: Initial intraoral examination**

Persistence of 62, 63 and 64 and absence of 22. A slight swelling was palpable in front of 62 during intraoral examination.

A panoramic radiography revealed a radiolucent image approximately 2 cm in diameter, homogeneous, unilocular, well-delimited, encompassing the crown of tooth 22 included, and pushing the teeth 23 posterosuperiorly towards the sinus floor and displacing the teeth 21 with apical mesiorotation and coronary distorotation (Figure 2). Three-dimensional imaging was subsequently carried out, revealing a hypodense lesion inserting itself into both the neck of 22 and 23 of approximately 21 mm x 17mm x 24 mm (Figure 3). The clinical and radiographic examinations were therefore in favor of a dentigerous follicular cyst of the teeth 22.
Large unilocular lesion, approximately 2 cm in diameter, located in the canine region. It is interesting to see that the cyst seems to be inserted both in the neck of the 22 in a low position and in the neck of the 23 in a superior-posterior position.

The temporary tooth 62, in direct communication with the cystic lesion, of this 9-year-old child was then extracted and a sample of the cystic membrane was obtained for histological analysis. A drain was placed in the cystic cavity through the alveolus of the 62 in order to decompress the cyst (Figure 4). Twice-daily rinsing with chlorhexidine for 2 weeks then with physiological serum of the cystic cavity was carried out by the patient, through the drain with a syringe.
FIGURE 4: Intra-oral situation 4 months after surgery.
The drain was placed at the site of maxillary left deciduous lateral incisor.

Anatomopathological analysis revealed a non-keratinized squamous epithelium of variable thickness containing inflammatory remodelling with polynuclear cells and lymphocytes, without any suspicious character, confirming the diagnosis of dentigerous cyst. Diagnoses of radicular cysts or potentially aggressive benign cysts such as ameloblastoma or keratocyst were therefore excluded.

In order to avoid mesialization of the left maxillary posterior sector, a Nance arch was placed with regular orthodontic appointments. After 4 months of cystic decompression, the drain was removed and teeth 63 and 64 were extracted. Clinical and radiographic follow-up, including dental panoramic radiography, were performed and showed progressive regression of the cyst and progression in coronary direction of the teeth 22 and 23 with root apexification (Figure 5).

<table>
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<th>Initial</th>
<th>+ 4 months</th>
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FIGURE 5: Radiographic follow up of the dentigerous cyst in the canine region.

The tooth 22 erupted progressively without further surgery. Orthodontic-surgical traction of tooth 23 was performed 1 year after the beginning of decompression and this tooth was functional after 3 years (Figure 6). Thus, thanks to a conservative approach over several years combining a minimal surgical technique by decompression and orthodontic treatment, the dentigerous cyst appeared to have been completely eliminated by the placement of the causal tooth 22 and the impacted tooth 23 (Figure 7).
FIGURE 6: Intra oral situation with orthodontic treatment at 4 years after surgery.

FIGURE 7: Three dimensional imagery 4 years after surgery.

Both 22 and 23 was on the arch and CBCT showed no radiolucent image around teeth, confirming cyst’s regression without any other surgery.
Discussion

The dentigerous cyst arises from the dental follicle of an unerupted or developing tooth, attached at the cemento-enamel junction. The frequency of odontogenic cysts in children is relatively low. It has been estimated that about 4%-9% of dentigerous cysts occur in the first decade of life [7]. Dentigerous cyst pathogenesis is related to pressure from an erupting tooth on the follicle that may obstruct circulation thus inducing to exudate accumulation between the reduced adamantin epithelium and the tooth crown [8]. The management of this lesion in children requires careful consideration and may be different from adults. In fact, when a cyst affects a child and causes delayed eruption, prognosis for the permanent teeth is often compromised, which is a source of anxiety for both the child and the parents. It is therefore interesting to be able to propose a reliable technique for cyst removal, while preserving the immature teeth involved.

According to Berretta et al [3], cystic decompression and marsupialization are both effective techniques in the management of benign cysts such as dentigerous and radiculodental cysts, but also more aggressive lesions such as keratocysts and unicystic ameloblastomas. Obviously, before considering these conservative techniques, it is important to make the correct diagnosis by anatomopathological sampling, as the prognosis of DC, keratocyst or ameloblastoma is not the same. Aggressive lesions such as ameloblastoma or keratocyst will inevitably require an additional procedure, whereas cystic decompression alone may be sufficient for a DC. In these different types of cyst, the use of decompression or marsupialization has shown no significant difference in cystic reduction [5]. Bone healing after cystic lesions removal is a multifactorial process depending on bone remodelling, the size of the lesion and the anatomical structures involved [9]. It is important to note that decompression appears to be effective in the case of DC, where resorption is more rapid than in radiculodental cysts or keratocysts [10-11]. This method has fewer complications than enucleation regarding to important anatomical structures preservation of and developing permanent tooth germs [12]. Although some cases report a risk of development of ameloblastoma in situ or micro-invasive ameloblastoma or other neoplastic transformations of the wall of the DC, it would appear that these decompression techniques are widely indicated in order to encourage the permanent teeth to remain on the arch [12].

Recently, the study by Nahajowski et al. [13] investigated the predictive factors favouring the placement on the arch of teeth impacted by a dentigerous cyst. It would appear that age and stage of dental development are predictive factors and correlate with spontaneous dental eruption after marsupialization. Spontaneous eruption occurs at the beginning of the first decade and when root development is less than or equal to half of complete formation [14-15]. This finding could be explained in part by the greater potential for bone regeneration in younger individuals. Tooth eruption does not always occur spontaneously after decompression or marsupialization, particularly when there is insufficient space to allow eruption or when no favourable axis is available [16-17], and may subsequently require additional orthodontic management to ensure optimal placement of the permanent teeth on the arch.

In our patient, the left maxillary lateral incisor (tooth 22) was at Nolla stage 7, while the canine (tooth 23) was at stage 6. His young age at the time of the fortuitous discovery of the cyst, the absence of complete root edification of the impacted teeth, and his compliance with treatment were key factors that had a major impact on the prognosis of the teeth, which are now functional. Decompression of the cyst lasted approximately 4 months. These results seem to be in agreement with those reported by Allon et al. [18], where the estimated mean decompression period was 7.5 months in children under 18 years of age. The review proposed by Berretta et al. [3] suggests a maximum time of 23.5 months. It is therefore essential to inform patients in advance of the time required for these techniques to be effective, in order to achieve optimum compliance, which is fundamental to the success of the treatment [13].

In this case, follow-up every two months was carried out by both an oral surgeon and an orthodontist, which enabled us to manage the placement of the permanent teeth in the arch as effectively as possible. Close collaboration is required to ensure that surgical traction of the canine is carried out at the right time, depending on its intraosseous three-dimensional position and the degree of root edification. It is also important to emphasise that no additional cystic enucleation surgery was used with complete recurrence of the cyst, confirmed by three-dimensional imaging. This result is also shared by Lizio et al. [19] who also suggested that simple cystic decompression could be a definitive treatment for dentigerous cysts without secondary surgical treatment, which was achieved in our patient. Long-term follow-up is still required however, recurrence of dentigerous cyst is seldom found, especially after complete removal of cyst or tooth eruption.

Conclusions

Given its prevalence, knowledge of the different treatments for dentigerous cysts, depending on the patient’s age and dental training, is fundamental. In children, a conservative approach by marsupialization or decompression will always be favoured in the first instance so as to encourage the maintenance on the arch of the permanent teeth affected by the cyst. Close collaboration between orthodontists and oral surgeons is essential.
Nevertheless, a histological analysis remains essential to avoid misdiagnosing cysts or more aggressive tumors such as keratocytes or ameloblastoma, which often require additional adjuvant surgery. Finally, in order to ensure that the treatment is effective, strict compliance and hygiene must be maintained throughout the course of treatment as a guarantee of success.

**Additional Information**

**Disclosures**

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**References**


