

CASE REPORT

Adapting treatment approaches for dentigerous cysts in paediatric and adult patients: A case series

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Abstract

Dentigerous cysts are common odontogenic cysts treated through surgical enucleation. This article presents paediatric and adult cases where surgical planning and treatment of dentigerous cysts were adapted according to various clinical challenges. Additionally, spontaneous regression of a dentigerous cyst associated with a maxillary canine is reported.

1 | BACKGROUND

Odontogenic cysts can be classified based on those which are inflammatory in origin such as the radicular and inflammatory collateral cyst and those which are developmental in origin.¹ The dentigerous cyst (synonymous with the term follicular cyst) is the most common type of odontogenic developmental cyst accounting for 24% of all cysts in the jaws.²

Its pathogenic mechanism is postulated to be related to tissue fluid accumulation from reduced enamel epithelial remnants causing expansion of the unerupted tooth follicle. Clinical presentation can be variable with most lesions remaining asymptomatic and progressively expanding until an infectious process initiates symptoms or the expansion of the lesion is recognized. Delayed or failure of eruption of teeth can lead to incidental discovery through radiological assessment.

Removal of the entire or part of the cystic lining is necessary with minimal delay to confirm diagnosis through histopathology and to exclude other lesions, which have similar clinical and radiographic appearances but require different

management strategies. For the majority of dentigerous cysts, treatment involves surgical removal of the cystic lining through enucleation with extraction or maintenance of the associated tooth. However, there are a number of different clinical presentations and numerous adapted treatment approaches for management of dentigerous cysts in both adult and paediatric patients. These encompass short and long-term treatment plans to preserve associated teeth and minimize risk of damage to vital structures.

We present a case series of three paediatric and two adult patients where the surgical treatment of their dentigerous cysts were modified according to their respective clinical challenges. Patient management and treatment were conducted at the University Dental Hospital of Manchester (UDHM). All patients had no relevant medical history and were followed up for a minimum of 6 months clinically and radiographically (unless otherwise specified). All cone beam computed tomography (CBCT) scans were undertaken using a low-dose paediatric protocol as per local guidelines.³ All patients or where appropriate, those with parental responsibility, provided consent for publication.

2 | PAEDIATRIC CASE REPORT

2.1 | Case 1

A 9-year-old boy was referred by his General Dental Practitioner (GDP) for a progressively enlarging, asymptomatic swelling focused on his right cheek. Clinical examination revealed a mixed dentition with multiple carious lesions, however, there was no maxillary expansion. Radiographic examination with an orthopantomogram (OPG) radiograph demonstrated a well-defined, corticated radiopacity associated with the unerupted permanent right maxillary canine (UR3) consistent with the appearance of a dentigerous cyst (Figure 1). The patient underwent extraction of the both maxillary deciduous canines and carious primary molars with restoration of the first permanent molars under local anaesthetic with the aid of inhalation sedation. It was anticipated that extraction of the carious deciduous canine would facilitate eruption of the permanent canine. The patient was subsequently reviewed nine months later and radiographic examination showed an improvement in the position of the unerupted UR3 with a reduction in the size of the lesion (Figure 2). A CBCT scan was undertaken which demonstrated the dimensional size of the cystic lesion enveloping the unerupted UR3 with bony expansion and encroachment of the right antral space, but no displacement or resorption of surrounding teeth.

A multi-disciplinary assessment with oral surgery and orthodontic specialists was undertaken and a decision made to marsupialize the cyst, following de-rotation and distalization of the maxillary right first premolar. This was deemed necessary to create space for an appliance to act as an obturator (Figure 3). On final review prior to surgery, a further radiograph (Figure 4) was taken which showed spontaneous resolution of the cystic lesion and bony infiltration. Therefore, surgical intervention was deemed to no longer be required and the patient placed on regular review.



FIGURE 1 An OPG radiograph demonstrating a well-defined, corticated radiopacity in the right maxillary antrum associated with the crown of the unerupted UR3 and extending from the UR6 to the UR2

2.2 | Case 2

A 7-year-old boy was referred by his GDP who noted the presence of supernumerary teeth as an incidental finding on a radiograph taken following dental trauma. On examination, both permanent maxillary central incisors (UR1 and UL1) were intact and demonstrated normal responses to sensibility testing. An upper standard occlusal radiograph revealed two tuberculate supernumerary teeth, palatally positioned in the anterior maxilla (Figure 5). Additionally, a well-defined, corticated radiolucency associated with the right supernumerary tooth was found. A CBCT scan was performed to provide further details of the lesion characteristics and to inform surgical planning (Figure 6).

The patient underwent surgical removal of the supernumerary teeth and enucleation of the cyst under general anaesthetic. Histopathological analysis confirmed diagnosis of a dentigerous cyst. A radiograph was taken during the 1-year review which showed resolution of the lesion and bony infiltration (Figure 7).

2.3 | Case 3

An 8-year-old boy was referred by his GDP regarding a persistent unilateral facial swelling which did not resolve following extraction of the suspected causative carious primary tooth. Clinical examination revealed buccal expansion of the maxillary canine region with associated displacement of the right maxillary canine and facial asymmetry (Figure 8). An OPG radiograph revealed a substantial, corticated radiopacity projected over the right maxillary sinus (Figure 9). This was further examined by CBCT which revealed a large cystic lesion associated with the unerupted permanent right maxillary canine (UR3) and second premolar (UR5) occupying almost the entirety of the maxillary sinus (Figure 10). Due to the extensive nature of the cyst (and dental caries affecting the right maxillary first permanent molar [UR6]), this patient was at high risk of losing several permanent teeth. The resulting consequence would be the patient only having his right maxillary central and lateral incisors and second permanent molar (UR1, UR2 and UR7 respectively) in the upper right quadrant with a high burden of future restorative treatment need. Therefore, the decision was taken to undertake marsupialization of the cyst under general anaesthesia with extraction of all carious maxillary primary molars and the carious maxillary right first permanent molar. In addition, a removable obturator appliance were designed and fitted immediately post operatively to facilitate cyst decompression (Figures 11 and 12).

The histopathological results confirmed the provisional diagnosis of a dentigerous cyst. The patient's facial asymmetry resolved and a subsequent radiographic follow-up demonstrated bony infiltration and positive migration of the UR3 and UR5 into the arch (Figure 13). The maxillary first



FIGURE 2 An OPG radiograph (A) demonstrating eruption of the UR3 in comparison to the initial radiograph (Figure 1). An upper standard occlusal radiograph (B) was also taken which highlights the size of the lesion in an antero-posterior plane and position of the UR4



FIGURE 3 Distalization and derotation of the UR4 with fixed appliances to create space for the UR3 in the arch and a post-surgery obturator

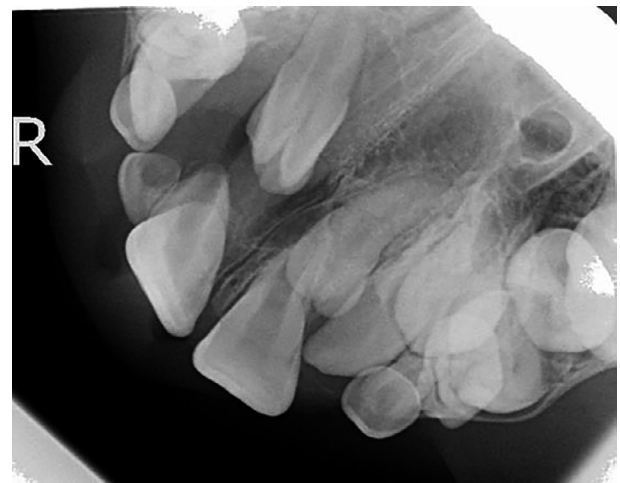


FIGURE 5 An upper standard occlusal radiograph revealed two unerupted tuberculate supernumerary teeth. There is a radiolucency associated with the right supernumerary tooth extending towards the developing right premolars and towards the midline



FIGURE 4 Pre-surgery OPG radiograph demonstrating spontaneous resolution of the cystic lesion associated with the UR3 and further improvement in its eruptive position

premolar (UR4) roots demonstrated arrested and incomplete root development, thus have a guarded long-term prognosis and the patient is currently under long-term review.

3 | ADULTS CASE SERIES

3.1 | Case 4

A 47-year-old male was referred to the UDHM by his GDP following an incidental finding of a asymptomatic radiolucency associated with an unerupted mandibular left third permanent molar (LL8). The patients' relevant medical history included smoking eight cigarettes per day.

Clinical examination were unremarkable with no swelling or altered sensation. A sectional OPG radiograph (Figure 14) revealed a well-defined and corticated radiolucency associated with the crown of a mesio-angularly impacted LL8. Additionally, the inferior alveolar nerve canal was deflected towards the cortex of the mandible. The occult nature of the lesion was evident as it had no associated clinical signs or symptoms. A CBCT scan was taken to fully examine the position of the inferior alveolar nerve canal and dimensions of the lesion. Its radiographic appearance was consistent with a dentigerous cyst, however, an odontogenic keratocyst or ameloblastoma could not be excluded.



FIGURE 6 Axial CBCT view showing the horizontal positions of the tuberculate teeth and their surrounding radiolucency



FIGURE 8 There is buccal expansion in the upper right maxillary canine region with a rotated UR4 in the canine space

Marsupialization was completed under local anaesthetic to facilitate cyst decompression. A biopsy sample confirmed the presence of an odontogenic cyst, which did not demonstrate features of an odontogenic keratocyst. A working diagnosis of a dentigerous cyst was maintained and the patient advised to flush the exposed cavity three times per day using a monojet syringe with saline. This was continued over a number of months to allow the cyst to decompress. The importance of compliance were discussed with the patient before undertaking this treatment plan.

The patient was reviewed during intervals and continued conservative management for 1 year. A sectional OPG radiograph was taken at the 1-year review (Figure 15) illustrating reduction in size of the cystic lesion, bony infiltration and improvement of the position of the deflected inferior alveolar nerve canal. The patient was consented for enucleation of the cyst and coronectomy or extraction of the LL8 under



FIGURE 7 One year review post-surgery showing resolution of the lesion with bony infiltration



FIGURE 9 An OPG radiograph revealed a corticated radiolucency in the right maxillary antrum extending to involve the unerupted UR3

general anaesthetic. During the procedure, the roots of the LL8 demonstrated mobility and the decision taken to extract the tooth. Post-operative review at the 2 month and 1 year period demonstrated no signs or symptoms of altered sensation and radiographically there was evidence of resolution of the cystic lesion with bony infiltration (Figure 16).

3.2 | Case 5

A 58-year-old female was referred by her GDP regarding a lesion associated with the maxillary left third permanent molar (UL8).

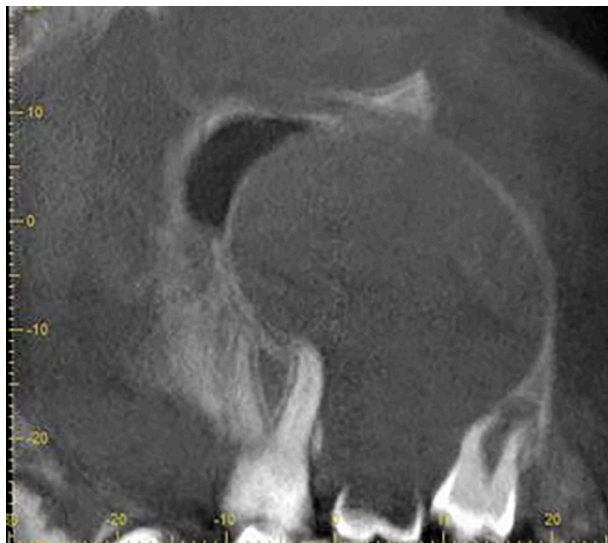


FIGURE 10 Sagittal CBCT scan view demonstrating a corticated cyst extending superiorly towards the orbital floor and medially towards the inferior meatus



FIGURE 11 Obturator appliance designed to facilitate cyst decompression was fitted immediately post-operatively

Clinical examination demonstrated a grossly carious maxillary left second permanent molar (UL7) and an unerupted UL8. Radiographically, the UL8 was inverted with an associated radiolucency (Figure 17). A provisional diagnosis of a dentigerous cyst was made, however an odontogenic keratocyst could not be excluded at this point. The patient underwent surgical extraction of the UL7 and UL8 with enucleation of the cyst under general anaesthesia. Perioperatively, an oral-antral communication was noted which was repaired with a buccal fat pad and buccal advancement flap. The histopathological report outlined features of an inflamed odontogenic squamous lined cyst however an odontogenic keratocyst could not be excluded. Regular clinical and radiographic review were undertaken to assess for recurrence and following no changes at the 6 month post-operative review (Figure 18), the patient was discharged to their GDP for continuation of monitoring.



FIGURE 12 View of the obturator appliance in situ



FIGURE 13 Post-operative radiograph taken during the review appointment showed resolution of the cyst and positive migration of the UR3 and UR5 towards the arch. Note the extensive root resorption of the UR4

4 | DISCUSSION

Dentigerous cysts are common odontogenic cysts of the jaws; being only second in incidence to the radicular cyst.⁴ The postulated pathological mechanism is accumulation of tissue fluid between the reduced enamel epithelium or the enamel organ, causing osmotic influx of fluid into the lesion and expansion of the follicle.^{5,6} The pressure of eruption or the breakdown of cells residing within the follicle due to obstructed eruption have been attributed to the increase in osmolality of cystic fluid, creating a gradient and initiating cyst expansion.^{5,7} Mechanisms have been proposed detailing non-vital deciduous teeth or their associated periapical pathosis causing secondary inflammation of the permanent successor follicle, initiating the formation of an inflammatory cyst.⁵ The cystic lining is an extension of the follicle hence is comprised of cuboidal or squamous epithelium.^{8,9}

As the fluid volume increases the lesion expands, however, they tend to be slow growing and asymptomatic in their early stages. Eventually, the lesion can result in clinically identifiable expansion or cause an infectious process



FIGURE 14 A sectional OPG revealed a well corticated radiolucency associated with the mesio-angularly impacted LL8. There is displacement and narrowing of the inferior alveolar nerve canal and the lesion extends towards the LL6



FIGURE 15 The 1 year post-marsupialization radiograph revealed reduction in size of the lesion with an improvement in the position of the inferior alveolar canal and bony infiltration around the LL6

initiating pain, which can often be the first presenting symptom. In addition, expansion of the lesion towards vital structures can manifest symptoms such as nerve paraesthesia.⁶



FIGURE 16 One year post-operative review following extraction of the LL8 demonstrates further bony infiltration



FIGURE 17 Initial radiographic assessment revealed the UL8 to be inverted with a corticated radiolucency associated with its unerupted crown. In addition, the UL7 has gross coronal caries

Mandibular third molars are the most likely teeth to be impacted and the most frequent site for occurrence of dentigerous cysts.^{10,11} The anterior maxilla is the second

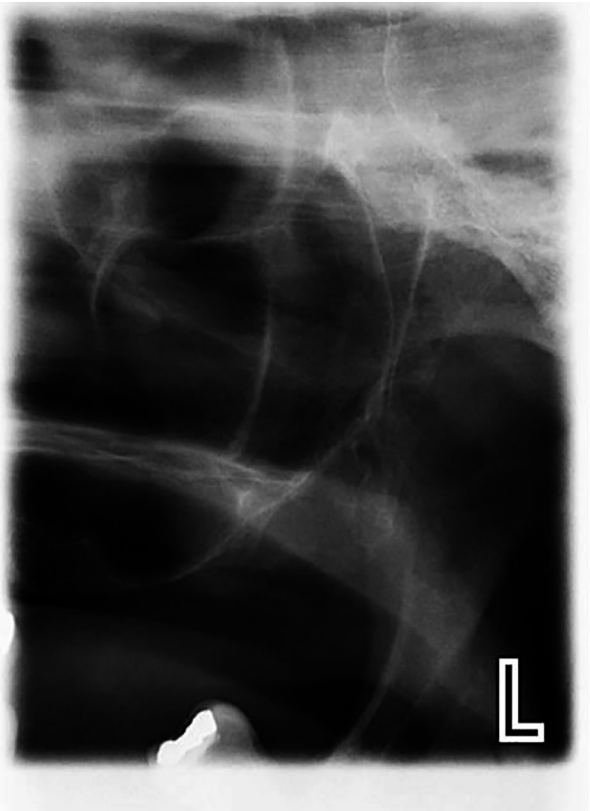


FIGURE 18 6 month post-operative radiographic review showed resolution of the cyst

most frequent site, attributed to the frequency of maxillary canine impaction and the presence of supernumerary teeth.¹²⁻¹⁴ Dentigerous cysts are associated with unerupted or impacted teeth and this explains the incidence pattern of disease manifestation mirroring the incidence of impacted teeth. Dentigerous cysts are more likely to arise in the mandible than maxilla, with reported rates of 70% and 30%, respectively.¹⁵

Radiological features demonstrate a well-defined unilocular radiolucency associated with the crown (specifically cemento-enamel junction) of an unerupted tooth. The slow expansive nature of the lesion can displace structures such as the mental foramen or inferior alveolar nerve canal. In addition, resorption of the apices of surrounding teeth can also be seen. Cross-sectional imaging through CBCT is recommended if there is uncertainty regarding the lesion's proximity to anatomical structures, if surgical planning for an extensive lesion is required or if it has invaded bony boundaries such as the cortex of the mandible or antral walls.¹⁶ Nonetheless, it is not possible to conclusively exclude other lesions of similar appearance such as the odontogenic keratocyst or ameloblastoma hence histopathological diagnosis is essential to rule out lesions that have similar clinical and radiological appearances.^{17,18}

Treatment involves removal of the cystic lining through enucleation and extraction or preservation of the associated tooth. Alternatively, marsupialization involves sectioning a bony window to facilitate decompression of the cyst and is

a method that can allow eruption of the associated tooth or reduction in size of the cyst. This is of particular importance in paediatric patients with a developing dentition where the burden of future dental treatment needs to be taken into consideration during treatment planning. Subsequent surgery may still be required to enucleate the entirety of the cystic lining but this two-staged approach can be useful to maintain teeth and improve separation of the cyst with vital structures.

Spontaneous regression of dentigerous cysts is considered rare and is seldom reported in literature.^{19,20} There were no cases identified in literature searches of dentigerous cysts associated with a maxillary canine spontaneously regressing. One case study reported regression of a dentigerous cyst associated with a mandibular canine on communication with the oral cavity, which has many similarities with case 1 and supporting the mechanism of tissue fluid being responsible for follicular expansion.¹⁹

Dentigerous cysts which are extensive in size, invade bony boundaries or affect vital structures may benefit from marsupialization to reduce the size of the cyst prior to enucleation. For child patients in the mixed dentition, a multidisciplinary approach can be helpful to fully treat their changing dentition and consider all implications of surgery. Every attempt should be made to minimize the invasiveness of surgery and preserve associated teeth.

5 | Consent to publish

All patients (or their respective guardian with parental responsibility for paediatric patients) provided written consent for publication.

CONFLICTS OF INTERESTS

The authors have no conflicts of interest to declare for this article.

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