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Dentigerous cyst of inflammatory origin: a case report

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ABSTRACT

Background

To document a case of a dentigerous cyst of inflammatory origin which developed due to periapical infection from a grossly carious deciduous tooth.

Methods

An 11-year-old male patient presented with a 4-month history of swelling within the right mandible. Radiological examination showed a cystic lesion in association with an impacted, displaced tooth 44 with an overlying carious 85. Marked expansion of the buccal bone was noted.

Results

Due to the special needs of this patient, radical treatment including extraction of tooth 85, enucleation of the cystic lesion as well as extraction of the impacted tooth 44 was performed.

Conclusion

Careful clinicopathological and radiological correlation of a large cystic lesion associated with an impacted tooth in the setting of an overlying grossly carious primary tooth will yield the diagnosis of a dentigerous cyst of inflammatory origin.

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Keywords

Dentigerous cyst, dentigerous cyst of inflammatory origin, follicular cyst, developmental odontogenic cyst

Conflict of interest

The authors declare they have no conflict of interest.

CASE REPORT

An 11-year-old male patient presented with a main complaint of swelling within the posterior right mandible. The patient stated he had been aware of the swelling for approximately four months. He was seen at a local clinic which provided him with antibiotics but no further treatment. The patient was referred to the Department of Operative Dentistry at Sefako Makgatho Health Sciences University after being on antibiotic therapy for more than a month with no resolution of the swelling. The patient provided a medical history of spastic cerebral palsy and autism for which he is being treated in the Department of Neurology. He is currently on Risperdal therapy.

On intra-oral examination, a grossly carious tooth 85 was noted with associated swelling that extended to involve the buccal cortical bone from tooth 84 to the mesial aspect of tooth 46. On palpation, the lesion was hard and rubbery. The patient exhibited marked drooling, the cause of which could not be distinguished between mental nerve impingement or due to the special needs condition of the patient. The initial clinical impression was that of a dental abscess due to the carious tooth 85. A panoramic radiograph subsequently showed a large cystic lesion on the lateral aspect of an impacted tooth 44. The 44 was displaced towards the inferior border of the mandible. The remaining dentition was crowded. There was distinct buccal expansion in association with the lesion which was discernible on panoramic radiograph in the lower right quadrant. The cyst had a well demarcated margin close to the impacted lateral surface of tooth 44 with blurring to an indistinct margin in the periapical region of tooth 85 (Figure 1).

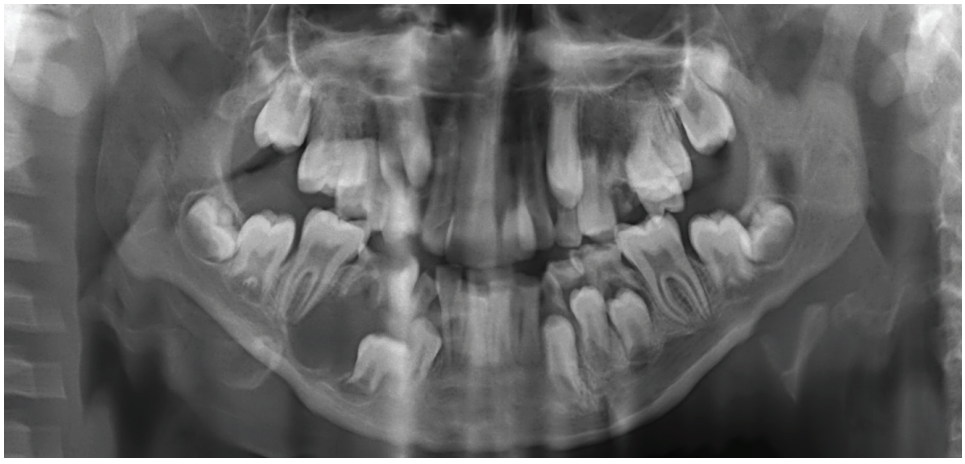

Figure 1: The panoramic radiograph obtained in this case depicts a grossly carious tooth 85 in association with a large lateral cystic lesion which is attached at the cemento-enamel junction of the impacted and displaced tooth 44. Generalised crowding of the remaining dentition is noted.

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DISCUSSION

Oral health care workers need to be able to distinguish between a wide variety of cystic lesions which occur in association with the crowns of teeth, some of which may represent neoplasms. A clinical differential diagnosis is formulated by intra-oral examination as well as inspection and evaluation of an appropriate radiographic image. Biopsy is advocated in many cases for histopathological analysis to reach the best diagnosis which directs treatment. The oral health care worker thus needs to have comprehensive knowledge of pathological lesions to reach a diagnosis and develop a treatment plan.

Dentigerous cysts (DCs) are common developmental odontogenic cysts which are attached to the tooth at the cemento-enamel junction (CEJ) (Figure 2). Any cystic lesion developing around the crown of an unerupted tooth may be termed a "follicular" cyst.[1] DCs develop as the erupting tooth applies pressure on the dental follicle which is often impeded by impacted teeth or where there is lack of intra-oral space. An unusual, unique variant of dentigerous cyst develops due to peri-apical infection from a carious deciduous tooth overlying the follicle. The inflammation stimulates the development of a cyst through induced replication of the reduced enamel epithelium (REE) which allows for fluid ingress and cyst development. These cysts often show hyperplastic epithelium in association with an inflammatory infiltrate.[2] The histopathological features of a typical dentigerous cyst include a low lining of stratified epithelium comprising cuboidal cells which resemble the REE surrounded by a fibrous cyst wall in which myxoid connective tissue changes are observed in association with inactive odontogenic cell rests.[3]

Figure 2: A gross morphological view of a dentigerous cyst which can be seen to attach at the cemento-enamel junction of the involved tooth. There are no luminal cyst contents.

Alternative theories as to the origin of a dentigerous cyst of inflammatory origin have been proposed by Altini and Benn. First, they suggest that a cyst forms within the follicle of the permanent tooth in the usual manner and then becomes secondarily infected by the periapical infection from the overlying carious deciduous tooth. Second, they propose that, on rare occasions, there may possibly be fusion of a radicular cyst located at the apices of the decayed deciduous tooth with the follicle of the unerupted permanent tooth. Last, it is suggested that the inflammatory exudate from the periapical infection associated with the apex of the carious deciduous tooth causes separation of the REE from the enamel with resultant cyst formation.[4]

Three radiographic varieties of dentigerous cyst have been documented. The most common is the central variety which presents as a well demarcated unilocular radiolucency surrounding the crown of an impacted tooth with attachment at the CEJ. The so-called lateral variant is most often seen in impacted third molar teeth where the cyst extends laterally along the root surface. This is commonly seen in mesio-angularly impacted teeth. The circumferential variant presents with the cyst surrounding the crown and roots of the tooth such that the tooth appears to be floating within the cystic space.[1] Although the majority of DCs are asymptomatic lesions which are usually identified coincidentally at check-up, it must be remembered that large DCs may often be multilocular and therefore may mimic a plethora of pathological lesions. DCs of the inflamed type present as radiolucent cystic lesions in association with an impacted tooth while above the lesion is a grossly necrotic retained deciduous tooth. In the event of large destructive lesions, incisional biopsy is advocated to exclude more sinister lesions and to confirm the diagnosis.[5]

DCs are common and are the second most frequently encountered odontogenic cyst after the radicular cyst.[6,7] The DC is, however, regarded as being developmental in nature while the radicular cyst is of inflammatory origin. DCs are estimated to comprise up to 20% of all odontogenic cysts.[2] Teeth most affected by DCs are the third molars and maxillary canines, followed by the second mandibular premolars, as these teeth are the last to erupt and most often the ones to encounter intra-oral space limitations.[6] A DC of inflammatory origin tends to show an overall predilection for occurrence within the mandible, generally in the premolar site and often in association with a grossly carious preceding primary molar tooth. They are also more frequent in male patients.[4] As a result, several clinical and radiological features are considered supportive of the diagnosis of a DC of inflammatory origin. These include younger age, location, symptoms, dental caries, cystic size, delayed eruption and impaction of permanent teeth.[6]

The treatment in most cases is radical and involves enucleation of the cyst together with extraction of the associated tooth. Other conservative strategies require a meticulous and accurate diagnosis and involve marsupialisation or decompression of the cyst with orthodontic movement of the associated tooth into place. This form of treatment is difficult, and success is best attained in children under the age of 10 years and where there is favourable depth of inclusion and germ angulation is less than 25º.[5] Extreme conservative management in such a case would include marsupialisation or decompression of the cyst wall in order to prevent neural fallout and the save the associated unerupted tooth which is then orthodontically moved into its normal position.[7,8] Such conservative management requires regular dental follow-up, daily local care and considerable co-operation from the patients and their parents.[5] Meticulous conservative therapy is not ideal in a patient with special needs. Although the definitive diagnosis of most cystic lesions requires histopathological analysis, clinical and radiological examinations have been shown to

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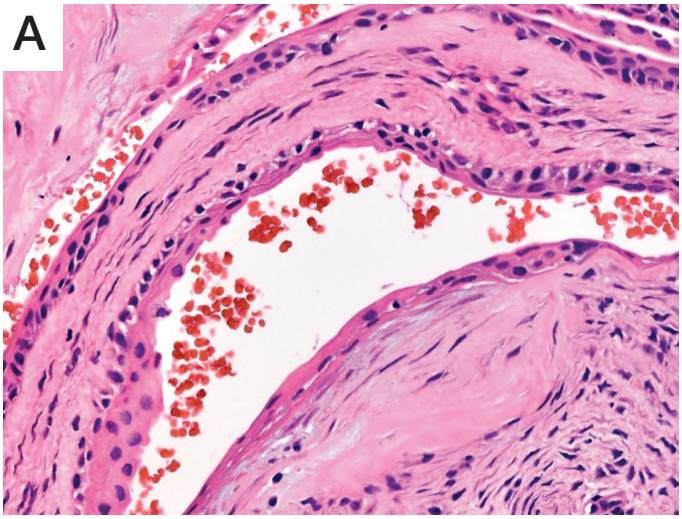
be of paramount importance in establishing a differential diagnosis and, in this case alone, was sufficient to dictate the immediate therapeutic intervention through enucleation and extraction of the two associated teeth, given the special needs of the patient.[8]

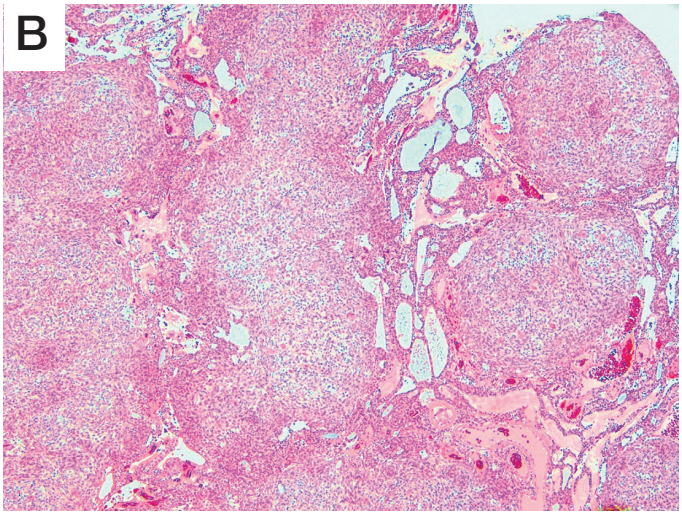
DCs, particularly in association with third molar teeth, should always be distinguished from hyperplastic follicles to avoid extraction of teeth which have the potential to erupt into their normal positions. A hyperplastic follicle and a DC may be distinguished from each other by means of radiology where the former measures up to 8mm, while a radiolucent lesion attached at the CEJ of the affected tooth measuring more than 8mm would be considered a DC.[8] On histology, a hyperplastic follicle and a DC are indistinguishable. Both will show a cystic lesion which is lined by a layer of uninflamed, uniform cuboidal cells of the REE (Figure 3A). Myxoid changes are frequent within the adjacent cyst wall as are occasional scattered quiescent odontogenic cell rests. Inflammation of an enlarging cyst may occur when there is breach of the lining or if a large cyst is biopsied which may introduce inflammation. Inflammatory changes may then alter the normal cyst lining which may become stratified and resemble that of a radicular cyst. The attachment of the cyst to the CEJ of the associated tooth will, however, assist with confirmation of DC.

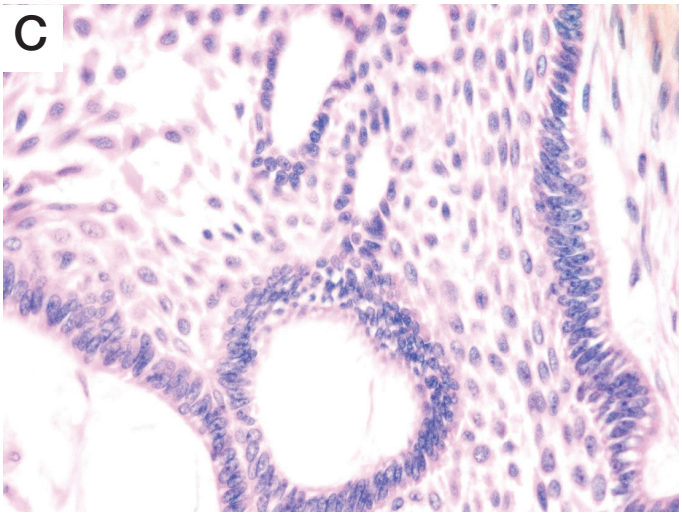
The pathogenesis of a DC has been alluded to. As the dental follicle enlarges to form a cyst, it becomes visible on radiograph as a well demarcated cystic lesion attached at the CEJ of the affected tooth.[2] There is often a need to distinguish this lesion from an early developing adenomatoid odontogenic tumour (AOT), a follicular odontogenic keratocyst (OKC) and a follicular ameloblastoma.[9-11]

An AOT in its earliest phases of development may mimic a DC; however, it should be noted that the cystic component often attaches lower than the CEJ on the root surface. Furthermore, if the AOT is more developed, the radiolucency surrounding the affected tooth may be seen to contain “snowflake radiopacities” within the lumen.[9] There is a high incidence of AOTs affecting the second mandibular premolar teeth as well as maxillary canines. Microscopically, an AOT is characterised by a large cystic space lined by a thick fibrous capsule. The odontogenic epithelial lining is arranged in sheets with duct-like spaces within whorls/rosettes.[9] (Figure 3B)

The follicular variants of the OKC and of ameloblastoma are virtually indistinguishable from a DC on radiology alone. Both an OKC and an ameloblastoma may develop from the dental follicle of any developing, erupting tooth resulting in the formation of







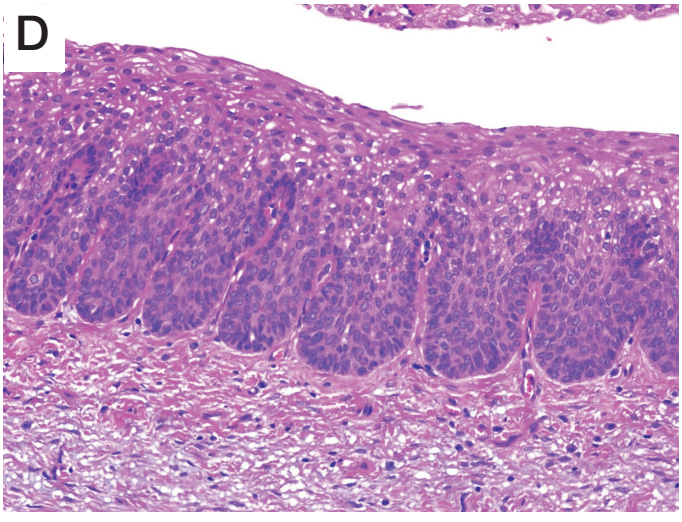


Figure 3A: The micrograph shows the histological features of an uninflamed dentigerous cyst in which the lining comprises a low stratified epithelium with many cuboidal cells which resemble the REE (Haematoxylin & Eosin; x20). Figure 3B: The micrograph shows a low-power image from part of an adenomatoid odontogenic tumour in which the epithelium is arranged in sheets with distinct peripheral whorls (Haematoxylin & Eosin; x10). Figure 3C: The characteristic features of an ameloblastoma are demonstrated in this micrograph. The basal cells are columnar with reverse nuclear polarisation, infranuclear vacuolation and subepithelial hyalinisation. A loosely textured luminal layer of stellate-like reticulum is prominently featured (Haematoxylin and Eosin; x40). Figure 3D: This micrograph shows part of an odontogenic keratocyst in which vague undulation of the surface epithelium is seen. This lesion shows pronounced basal budding (Haematoxylin & Eosin, x40).

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a radiolucent lesion surrounding the crown of the tooth resembling a DC. These lesions have the potential to increase to large sizes at which time there may be associated cortical expansion or even destruction. Most DCs are slow-growing, asymptomatic lesions detected as incidental findings at check-up on a routine panoramic radiograph. Cysts that enlarge and go on to cause cortical bone expansion or breakthrough may be felt as fluctuant lesions. Large DCs have the potential to cause bone destruction and displacement of neighbouring teeth. They may even prevent the eruption of adjacent teeth.[5]

It is always advised to biopsy large cysts to rule out diagnoses such as OKCs and ameloblastomas for the most appropriate therapeutic intervention. An ameloblastoma on histology will show a lining of distinct columnar basal cells with reverse nuclear polarisation and infranuclear vacuolation. Prominent underlying hyalinisation is a frequent feature. The more luminal cells are loosely textured and resemble the stellate reticulum of the dental organ (Figure 3C). An OKC, by contrast, is histologically characterised by a uniform layer of stratified squamous cells, usually six to eight cells in thickness. The surface of the cells has a corrugated, undulated appearance. The junction of the lining epithelium with the underlying cyst wall is flat, allowing for easy separation. Basal budding or daughter cyst formation may be seen, all of which increase the probability of recurrence (Figure 3D). It is thus the accepted treatment of choice of large follicular cystic lesions to be enucleated together with extraction of the associated tooth. Depending on the diagnosis, curettage, resection or the use of Carnoyl's solution is proposed.

Patients with spastic cerebral palsy and autism tend to have poor oral hygiene. As a result of damage to the brain before, during or after birth, patients with spastic cerebral palsy often exhibit jerky, uncoordinated movements, muscle tightness and joint stiffness.[12-14] Children with these disorders may be exceptionally difficult to treat in the dental chair. The treatment as was performed in this case is thus appropriate and careful clinical follow-up is advised, particularly with regard to the maintenance of oral hygiene. The side effects of Risperdal as prescribed for this patient includes enhanced difficulty and control of joint movements as well as excessive drooling, which further hinders more conservative management.[15]

CONCLUSION

Most dentigerous cysts are developmental in origin. In young children, however, there may be a unique situation in which a dentigerous cyst develops because of periapical infection from a necrotic overlying deciduous tooth, as demonstrated in this case report. Furthermore, the patient in this case is afflicted with both autism and spastic cerebral palsy and has been prescribed a psychotropic drug which limits the clinical management. The pathogenesis of a dentigerous cyst because of adjacent inflammation remains a point of interest.

REFERENCES

1. Hasan S, Ahmed SA, Reddy LB. Dentigerous cyst in association with impacted inverted mesiodens: Report of a rare case with a brief review of literature. Int J Appl Basic Med Res. Sep 2014;4(Suppl 1):S61-64
2. McKinney SL, Lukes SM. Dentigerous cyst in a young child: a case report. Can J Dent Hyg. Oct 2021;55(3):177-181
3. Sohn C RJ, Nam I, Shin S, Lee J. Cystic lesion between a deciduous tooth and the succeeding permanent tooth: a retrospective analysis of 87 cases. J Korean Assoc Oral Maxillofac Surg. 2022;48(6):342-347
4. Benn A, Altini M. Dentigerous cysts of inflammatory origin. A clinicopathologic study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. Feb 1996;81(2):203-209
5. Khalifa C, Garma M, Mabrouk R, Slim A, Bouguezzi A, Selmi J. Conservative management of dentigerous cyst in children: Report of two clinical cases. Clin Case Rep. Apr 2023;11(4):e7051
6. Hye Joo Son YJJ, Jin Sook Jeong, Do-Young Kang. An inflammatory dentigerous cyst shows rim uptake on bone scan: a case report. Nucl Med Mol Imaging. 2014;48(1):79-81
7. Antunes D, Albisetti A, Fricain M, Cherqui A, Derruau S. Management of Permanent Teeth in Dentigerous Cysts in Children: A Case Report. Cureus. Aug 2023;15(8):e44062
8. Perez A, Lenoir V, Lombardi T. Dentigerous Cysts with Diverse Radiological Presentation Highlighting Diagnostic Challenges. Diagnostics (Basel). Aug 19 2022;12(8)
9. Sethi S KM, Aggarwal P, Indra Kumar HS, Sugandhi CD, Singh S. A case report and short review of changing trends in the site of occurrence of adenomatoid odontogenic tumour: Unravelling the past 15 years. Dent Res J. 2016;13:462-471
10. Hadziabdic N, Dzinovic E, Udovicic-Gagula D, et al. Nonsyndromic Examples of Odontogenic Keratocysts: Presentation of Interesting Cases with a Literature Review. Case Rep Dent. 2019;2019:9498202
11. Meshram M SL, Dhuvad J, Anchlia S, Vyas S, Shah H. Conservative Management of Unicystic Ameloblastoma in Young Patients: A Prospective Single-Center Trial and Review of Literature. J Maxillofac Oral Surg. 2017;16(3):333-341
12. Delli K, Reichart PA, Bornstein MM, Livas C. Management of children with autism spectrum disorder in the dental setting: concerns, behavioural approaches and recommendations. Med Oral Patol Oral Cir Bucal. Nov 1 2013;18(6):e862-868
13. Jan BM, Jan MM. Dental health of children with cerebral palsy. Neurosciences (Riyadh). Oct 2016;21(4):314-318
14. Zerman N ZF, Chirumbolo S, Zangani A, Mauro G, Zoccante L. Insights on dental care management and prevention in children with autism spectrum disorder (ASD). What is new? Front Oral Health. 2022;27(3):998831
15. Becker DE. Psychotropic drugs: implications for dental practice. Anesth Prog. Fall 2008;55(3):89-99

CPD questionnaire on page 346

The Continuing Professional Development (CPD) section provides for twenty general questions and five ethics questions. The section provides members with a valuable source of CPD points whilst also achieving the objective of CPD, to assure continuing education. The importance of continuing professional development should not be underestimated, it is a career-long obligation for practicing professionals.