Dentigerous Cyst Mimicking Odontogenic Keratocyst: A Rare Case Report with 12 Month Follow up

Rose M Joseph[1], Subhagatha Chakraborty[2], Anupama Nayak P[3], Dharnappa Poojary[4], Ashwin Rao [5], Nandita KP [6], Karuna YM[7]

[1]Former Post Graduate Student, Department of Pediatric and Preventive Dentistry, [2]Former Post Graduate Student, Department of Oral and Maxillofacial Surgery, [3]Reader, Department of Pediatrics and Preventive Dentistry, [4]Associate Professor, Department of Oral and Maxillofacial Surgery, [5]Associate Professor, Department of Pediatric and Preventive Dentistry, [6]Associate Professor, Department of Oral Pathology and Microbiology,[7]Reader, Department of Pediatric and Preventive Dentistry, Manipal College of Dental Sciences, Mangalore, Manipal Academy of Higher Education, Manipal, Karnataka, India, 576104

Abstract

Dentigerous cyst is one of the most commonly seen odontogenic cyst in children. It is characterized by a unilocular radiolucent lesion that encloses permanent tooth buds. Buccal bony expansion and a missing tooth is the most common clinical feature. Various treatment modalities have been mentioned in the literature for management of dentigerous cysts. This case report presents the management of a left mandibular dentigerous cyst in a 10-year-old boy by surgical enucleation and its follow up.

***Key Words**: Dentigerous Cyst, Mandible, Molar.*

Introduction

A dentigerous cyst is a well-defined, odontogenic lesion that surrounds the crown of an unerupted tooth arising from the epithelial remnants of the tooth forming organ.[1] It can be developmental or inflammatory in origin and is usually asymptomatic unless infected. [2] In children and adolescent, increased incidence of dentigerous cysts and Keratocystic Odontogenic Tumour (KCOT) are found when compared to other pathologies. [3] Dentigerous cyst is managed by enucleation and marsupialization.[4] In this case report we present a case of unilateral dentigerous cyst resembling a KCOT in a 10 year old boy with its management and a 12 month follow up.

Corresponding author:
Dr Anupama Nayak P, MDS,
Reader, Department of Pediatric and Preventive Dentistry, Manipal College of Dental Sciences, Mangalore, Manipal Academy of Higher Education, Manipal, Karnataka, India, Light House Hill Road-576104. Mobile no:+919945923865
Email id: anupama.np@manipal.edu

Case Report

A 10-year-old male boy had visited the Department of Pediatric dentistry with a swelling since 3 days in relation to lower left back region of face. No history of pain, fever or other systemic symptoms were present. On extra oral examination (Fig 1A), there was a hard localized unilateral swelling of size 1.5x1.5cm on the lower left lateral aspect of cheeks inferior & distal to the lip commissure with no surface redness or increase in surface temperature. Intra oral examination (Fig 1B) showed restored lower left second primary molar with grade II mobility but no tenderness on percussion. There was a localized bony hard non tender swelling of the buccal and lingual gingival region extending from distal aspect of 73 to the mesial aspect of 36 with obliteration of buccal vestibule and expansion of the buccal and lingual cortical plates.

A panoramic radiograph (Fig 1C) showed endodontically treated 75 with complete root resorption and a unilocular well defined oval radiolucency measuring 2x1.7cm in relation to the tooth bud of 35 extending from the mesial aspect of 34 to the mesial aspect of 36. Based on the clinical and radiographic

findings, a provisional diagnosis of dentigerous cyst was made.

To evaluate the lesion three dimensionally a CBCT scan was advised which (Fig 1D,1E, 1F) showed a well-defined radiolucency measuring 17.85x20.63x18.25 extending from the periapical region of 34 to 36 and surrounding the crown of developing 35. There was expansion of buccal and lingual cortical plates with thinning of the plates and partial loss of buccal cortical plate along with the root resorption of 75. Based on the above findings from CBCT a diagnosis of Keratocystic odontogenic tumour (KCOT) was given.

A confirmed diagnosis of the pathology can be obtained only after histological examination. Management of KCOT requires a more aggressive approach with marginal bone resection [5] along with complete removal of the lesion when compared to that of dentigerous cyst wherein enucleation [2] is the standard approach. Considering these factors and the age of the child, surgical enucleation of the cyst under general anesthesia (GA) was planned after consulting with the Department of Oral and Maxillofacial Surgery and after obtaining informed consent from the parent.

Extraction of 75 was done following which buccal and lingual flaps were raised to visualize the lesion under General anesthesia. Part of the thin and friable buccal cortical plate was removed following which the entire cystic lesion was enucleated (Fig 1G,1H,1I). As the cyst was firmly attached to the neck of the developing 35 it was decided to extract it. Following saline irrigation the flaps were closed with non resorbable sutures. The biopsy specimen was sent for histopathologic examination.

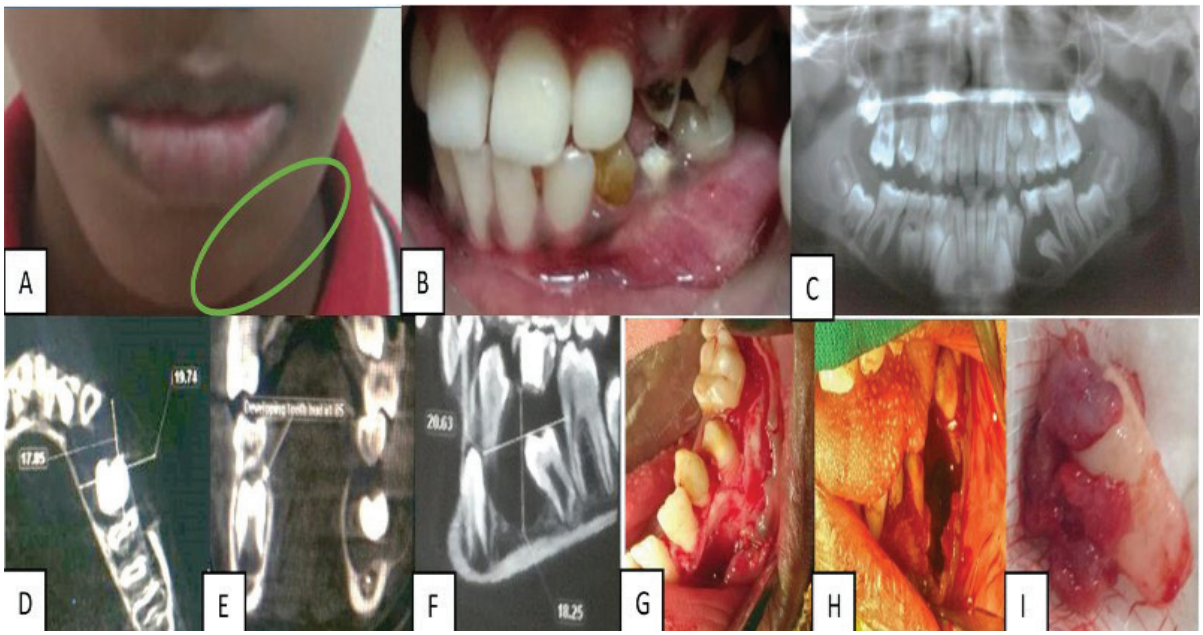

Fig 1: (A) Extra oral swelling on the left side. (B) Left buccal vestibule. (C) Orthopantomogram (D, E and F) CBCT (G) Intraoral vestibular incision (H and I) Removal of second deciduous molar with cyst.

Histopathologic evaluation revealed cystic lining of 2-3 cell layers of non-keratinized stratified squamous epithelium with fibro cellular connective tissue capsule which was attached to the cemento enamel junction of the tooth giving a confirmed diagnosis of dentigerous cyst (Fig 2A,2B).

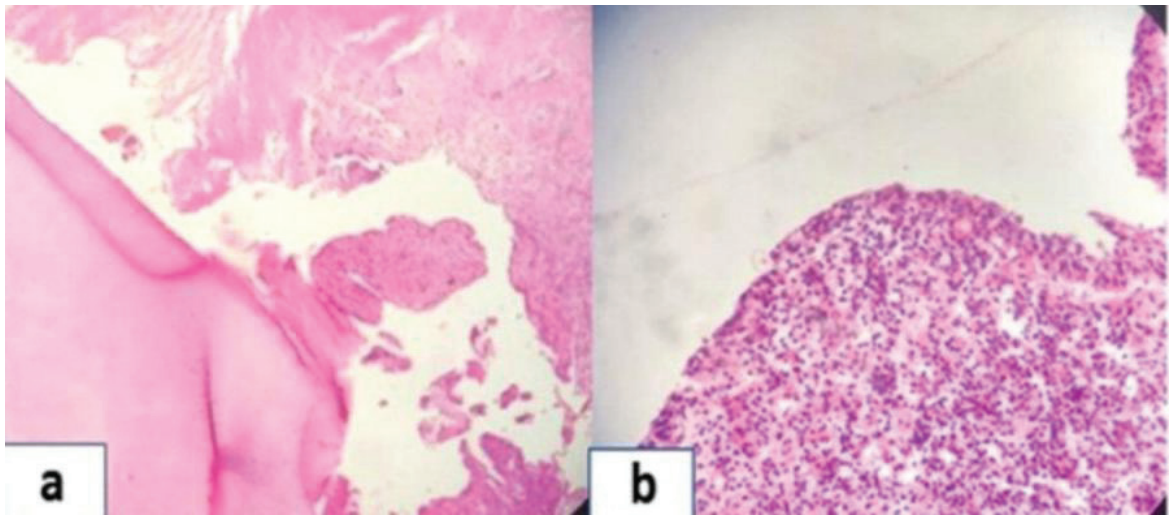

Fig 2: (A) Cystic lining attached to the CEJ (B) Stratified squamous epithelium lining the wall.

Patient was recalled after 1 week for suture removal and follow up visits at 1, 3, 6 & 12 months. During follow up visits, (Fig 3A,3B) uneventful healing of the lesion was noted.

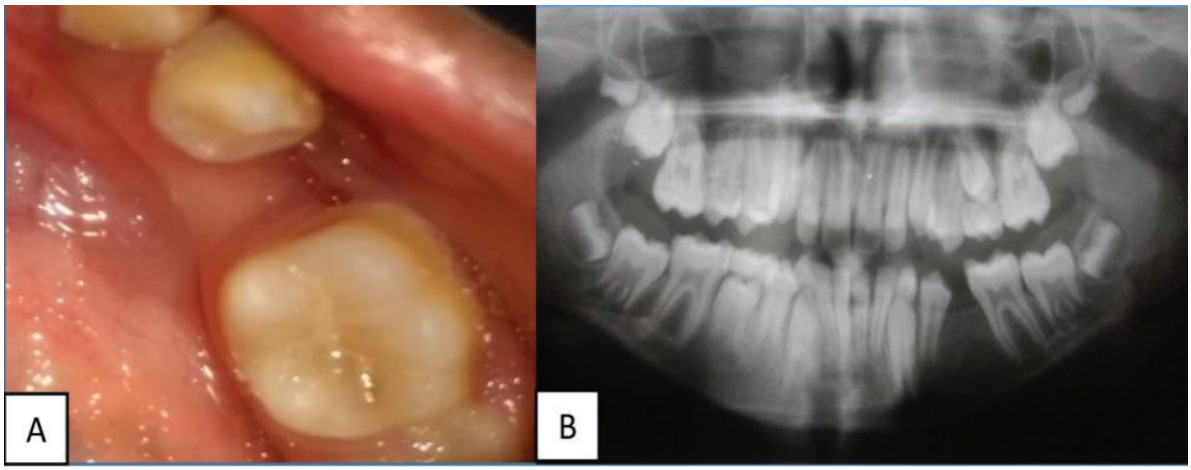

Fig 3: (A and B) Post op photograph and OPG at 12 months.

Discussion

Odontogenic cysts are among the commonest lesions encountered in children.[5,6] According to Manor et al[7] among the cystic lesions seen in children, developmental cysts were found at higher rate. A prevalence study by Li et al[3] on the occurrence of developmental cysts in children showed that 97.8% of the encountered lesions were dentigerous cyst and Keratocystic Odontogenic Tumour (KCOT). They also found that an increased risk of occurrence of KCOT as compared to dentigerous cyst in adolescent. This is in accordance with Tkaczuk et al[8] wherein out of 57 patients with true cystic lesions 33% were keratocystic odontogenic tumors and 30% were dentigerous cysts.

In children a dentigerous cyst may arise from an infected carious predecessor tooth or due to the continuous irritation of the follicle of the permanent tooth bud by obturating material of the treated primary tooth.[2] In the present case dentigerous cyst was of inflammatory origin developed from the pulpectomy treated deciduous second molar. Management of dentigerous cysts is commonly done by surgical enucleation and curettage with removal of the impacted tooth or teeth. However, if the lesion has larger dimensions and is causing tooth displacement and involves multiple teeth with extensive loss of bone marsupialization or decompression,

followed by enucleation later on can be attempted.[2]

In the present case, there was mixed opinion regarding diagnosis of the cyst suggested from OPG and CBCT, but considering age of the patient, enucleation of cyst was done with extraction of the associated tooth. On surgical exposure the cystic lining was fibrous and thick as opposed to the thin friable lining usually seen associated with KCOT. The lesion was removed as a whole which is difficult in case of KCOT. Extraction of developing second premolar was done as the lesion was found to be firmly attached surrounding the crown. The biopsy report confirmed the lesion as a dentigerous cyst due to presence of fibrous capsule that was attached at the CEJ which is characteristic of dentigerous cyst.[1]

Conclusion

Management of odontogenic lesions like dentigerous cyst in children can be challenging. Thus it is important to understand the nature of such lesions thoroughly backed by good clinical history and radiographic aids which help the clinicians to arrive at proper treatment protocol.

Conflict of Interest: No conflict of interest present.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parents have given their consent for images and other clinical information of the patient to be reported in the journal. The patient's parents understand that the name and initials of the patient will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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References

1. Devi P, Thimmarasa VB, Mehrotra V, Agarwal M. Multiple dentigerous cysts: a case report and review. J Maxillofac Oral Surg 2015;14:47–51.
2. Kumar R, Singh RK, Pandey RK, Mohammad S, Ram H. Inflammatory dentigerous cyst in a ten-year-old child. Natl J Maxillofac Surg 2012;3:80-3.
3. Li N, Gao X, Xu Z, Chen Z, Zhu L, Wang J et al. Prevalence of developmental odontogenic cysts in children and adolescents with emphasis on dentigerous cyst and odontogenic keratocyst (keratocystic odontogenic tumor). Acta Odontol Scand 2014;72: 795-800.
4. Arjona-Amo M, Serrera-Figallo MA, Hernández-Guisado JM, GutiérrezPérez JL, Torres-Lagares D. Conservative management of dentigerous cysts in children. J Clin Exp Dent 2015;7: 671-4.
5. Ahmed HK, Bassel Tarakji. Odontogenic Keratocyst in Children: A Review. Open Dent J 2016;10:117-23.
6. Prosdócimo ML, Agostini M, Romañach MJ, de Andrade BA. A retrospective analysis of oral and maxillofacial pathology in a pediatric population from Rio de Janeiro-Brazil over a 75-year period. Med Oral Patol Oral Cir Bucal 2018;23:e511-e517.
7. Manor E, Kachko L, Puterman MB, Szabo G, Bodner L. Cystic lesions of the jaws- a clinicopathological study of 322 cases and review of the literature. Int J Med Sci 2012;9: 20-6.
8. Tkaczuk AT, Bhatti M, Caccamese JF Jr, Ord RA, Pereira KD. Cystic Lesions of the Jaw in Children: A 15-Year Experience. JAMA Otolaryngol Head Neck Surg 2015;141: 834-9.