

Dentigerous Cyst Associated with a Deciduous Tooth: Report of a Case and Review of the Literature

Case Report

Najme Anbiaee¹, Maryam Rajabi², Hamed Ebrahimnejad³

¹ Associate Professor, Department of Oral and Maxillofacial Radiology, Maxillofacial Diseases Research Center, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.

² Postgraduate student of oral and maxillofacial Radiology, Department of Oral and Maxillofacial Radiology, Maxillofacial Diseases Research Center, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.

³ DDS, MSc, Oral and Maxillofacial Radiologist, Department of Oral and Maxillofacial Radiology, School of Dentistry, Kerman University of Medical Sciences, Kerman, Iran.

Received: Feb 15, 2016

Accepted: Jul 23, 2016

Corresponding Author:

Maryam Rajabi

Address:

Oral & Maxillofacial Radiology Department, Faculty of Dentistry, Mashhad University of Medical Sciences, PO Box: 91735-984, Mashhad, Iran.

Telephone: +98-51-38829501

Email: Dr.mr.rajabi@Gmail.com

Fax: +98-51-38829500

Abstract

Introduction:

Dentigerous cyst is a benign developmental lesion of the jaw. It is most commonly occurs during the second and third decades of life and has rarely been reported in association with a deciduous tooth. We report a case of two-year old girl who presented with an unerupted central incisor. According to the radiographic findings, she was diagnosed with a dentigerous cyst and underwent surgical enucleation. The final diagnosis was confirmed by histopathological analysis. We briefly discussed the characteristics of similar cases.

Key words:

•Incisor• Deciduous• Dentigerous Cyst

Case Report

In May 2009, a two-year seven-month old girl was presented to our dental clinic with the chief complaint of an unerupted lower incisor. She was born from a triplet pregnancy with healthy siblings. Her past medical and dental histories were unremarkable and she appeared healthy with normal development. Her extra- and intra-oral examinations were normal, except for an absent lower left central incisor (Figure 1). There was no mucosal swelling or redness in the region. The adjacent incisors had erupted and their alignment appeared normal. Radiographic examination of the region revealed a well-defined, unilocular radiolucency surrounding the crown of the unerupted central incisor (Figure 2). Based on the clinical examination and radiographic findings, the patient was initially diagnosed with dentigerous cyst (DC) and surgical enucleation of the lesion was advised. However, her parents refused the surgery and the patient was followed for two years. In the follow-up visits, there was no new complaint and the extra- and intra-oral examinations were the same as on the initial visit.

A follow-up radiographic examination was conducted, revealing a calcified fleck adjacent to the lesion (Figure 3). According to the radiographical findings, the differential diagnoses were DC, calcifying odontogenic cyst and odontoma. In June 2011, surgical enucleation was discussed with the parents and they consented to the treatment. The patient underwent surgery under general anesthesia and the cyst, along with the associated tooth, were removed and sent for histopathologic examination.

In gross examination, the specimen measured 9*9*4 mm³ and consisted in a cream-yellow tissue with soft texture, attached to the tooth. Microscopic examination of the hematoxylin and eosin stained sections revealed a cystic lumen with a thin layer of non-keratinized odontogenic epithelium and a fibromyxoid stroma (Figure 4). No evidence of inflammation or calcified material was noted and the diagnosis of DC was confirmed. The short-term post-operative period was uneventful. Unfortunately, the patient was lost in the long-term follow-up period. At the final follow-up visit, we obtained written informed consent from the parents of the child.



Figure 1. The intra-oral view, showing the missing deciduous central incisor and normal mucosa without noticeable expansion.



Figure 2. Periapical radiograph of the mandibular incisors, showing a well-defined, corticated pericoronal radiolucency associated with the left central incisor.



Figure 3. The two-year follow-up radiograph showed no explicit expansion. Small opacity was present on the lateral wall of the lesion.

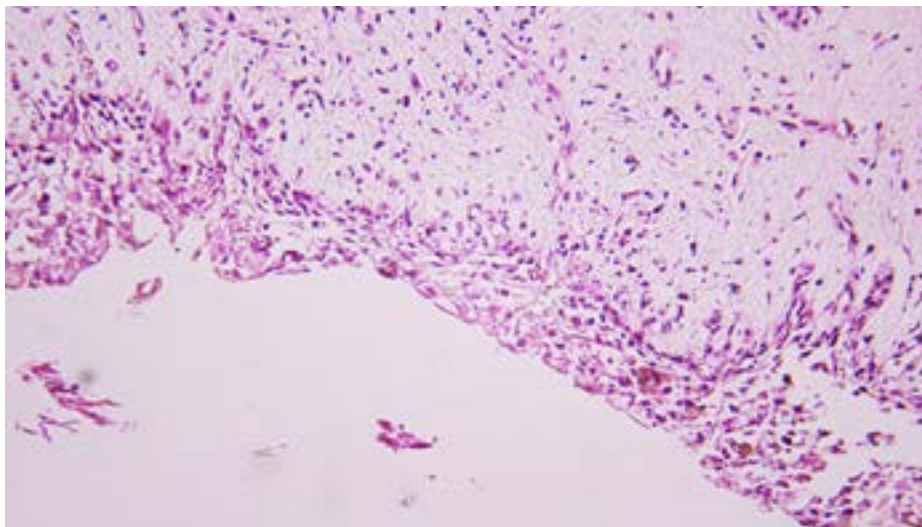


Figure 4. Photomicrograph of the lesion showing the odontogenic epithelial lining with adjacent loose connective tissue (H & E, original magnification, x100).

Discussion

Dentigerous cyst (DC) is one of the most common odontogenic pathologies of the jaw and accounts for nearly 20.1% of these lesions.⁽¹⁾ DC is most commonly associated with mandibular third molar.^(2,3) Despite the developmental origin of DC, its occurrence in association with deciduous teeth is extremely rare. Here we report a case of DC associated with an unerupted deciduous incisor in a two-year old girl.

DCs are extremely rare during the first decade of life and most reports have shown that patients in this age group account for less than 10% of all cases.^(4,5) The presented patient was unique for her age in the association of the lesion with a deciduous tooth and the location of the cyst.

There is an extensive body of literature concerning the pathogenesis, diagnosis and treatment of DC associated with permanent teeth. However, research on DC in deciduous teeth is limited to a

few case reports.

In the review of the literature, we found nine pediatric case reports of DC in association with deciduous teeth (Table 1).⁽⁶⁻¹⁴⁾ Unerupted tooth and painless swelling were the most common presentations of DC among the reported cases. To the best of our knowledge, the youngest reported case was a six-week old female who presented with a maxillary swelling during early days of infancy and was diagnosed with DC associated with the supernumerary maxillary left lateral incisor.⁽⁷⁾ DC of the deciduous tooth has also been reported in association with a number of other conditions, including fetal alcohol syndrome and odontoma.^(11,13) Also of note, deciduous molars were the most commonly-affected teeth in abovementioned case reports. Accordingly, our case is one of the youngest reports of DC associated with a deciduous tooth and the only one in association with a mandibular incisor.

Table 1. Characteristics of the reported cases of dentigerous cyst associated with a deciduous tooth

Author (year)	Gender	Age	Presentation	Associated condition	Associated deciduous tooth	treatment
Our case	Female	2 years, 7 months	Unerupted tooth	-	Mandibular left central incisor	Enucleation
Morishita et al. (2010) ⁶	Male	2 years, 9 months	Unerupted tooth, maxillary swelling	-	Maxillary 2nd molar	Marsupialization
Vucicevic Boras et al. (2007) ⁷	Male	6 weeks	Maxillary swelling	Supernumerary tooth	Maxillary left lateral incisor	Observation
Nukata et al. (1999) ⁸	Male	4 year, 11 months	Unerupted tooth	-	Mandibular 2nd molar	Enucleation
Miki et al. (1996) ⁹	Male	1 year, 9 months	Unerupted tooth, swelling	-	Mandibular left canine	Marsupialization
Kusukawa et al. (1992) ¹⁰	Male	2 years	Unerupted tooth, swelling	-	Maxillary 2nd molar	Marsupialization
Motokawa et al. (1990) ¹¹	Female	3 years	unerupted tooth	Odontoma	Maxillary 2nd molar	Marsupialization
Kishimoto et al. (1988) ¹²	Male	5 months	Mandibular swelling	-	Mandibular 1st molar	Marsupialization
Rosenlicht et al. (1979) ¹³	Male	8 months	Mandibular swelling	Fetal alcohol syndrome, several maxillofacial anomalies	Mandibular molar	Marsupialization
Yamanaka et al. (1977) ¹⁴	Male	7 year	Unerupted tooth, mandibular swelling	Past history of facial trauma	Mandibular 2nd molar	Enucleation

In approach to the patients with DC, plain radiography is often sufficient for the initial diagnosis. As of the present case, the typical radiographic presentation of DC is a well-defined unilocular cyst associated with the crown of an unerupted tooth.^(2, 3, 15) However, there are other lesions, such as keratocystic odontogenic tumor and unilocular ameloblastoma, which may resemble the radiographic appearance of DC.⁽¹⁶⁾

Accordingly, despite the critical role of imaging studies, histopathologic examination of the lesion is necessary to confirm this diagnosis. In the present case, there was a small radio-opacity adjacent to the lesion in the follow-up radiograph. Considering the absence of calcification or other lesions in the histopathologic examination, this opacity is likely due to a dense bony trabeculation.

Conclusion

In conclusion, despite the low incidence of DC in early childhood, it should be considered as a potential diagnosis in patients with unerupted tooth, since early treatment is necessary to avoid complications.

References

1. Johnson NR, Gannon OM, Savage NW, Batstone MD. Frequency of odontogenic cysts and tumors: a systematic review. *J Investig Clin Dent*. 2014;5:9-14.
2. Som PM, Curtin HD. *Head and Neck Imaging*. 5th Ed. US: Elsevier Health Sciences; 2011. p. 1476-1478.
3. Larheim TA, Westesson P-LA. *Maxillofacial Imaging*. New York: Springer Science & Business Media; 2006. p. 30.
4. Ledesma-Montes C, Hernández-Guerrero JC, Garcés-Ortíz M. Clinico-pathologic study of odontogenic cysts in a Mexican sample population. *Arch Med Res*. 2000;31:373-376.
5. Shear M, Speight P. *Cysts of the Oral and Maxillofacial Regions*. 4th Ed. New Jersey: John Wiley & Sons; 2007. p. 59-60.
6. Hiroshi M, Katsuko H, Motohiro G, Ryosuke K, Masahiro N, Kenji K. A case of dentigerous cyst associated with a deciduous tooth in a 2-year-old child. *Jpn J Oral Maxillofac Surg*. 2010;56:380-383 (in Japanese, abstract in English).
7. Vucicevic Boras V, Mohamad Zaini Z, Savage NW. Supernumerary tooth with associated dentigerous cyst in an infant. A case report and review of differential diagnosis. *Aust Dent J*. 2007;52:150-153.
8. Nukata J-i, Kodama Y, Matsuyama H, Matsumura T, Sakuda M, Kishino M. A case of follicular cyst associated with a deciduous molar. *Jpn J Oral Maxillofac Surg*. 1999;45:360-362 (in Japanese, abstract in English).
9. Miki T, Tachikawa N, Oda M, Kurachi T, Sakai E, Enomoto S. A case of a dentigerous cyst arising in a deciduous tooth. *Jpn J Oral Maxillofac Surg*. 1996;42:206-208 (in Japanese, abstract in English).
10. Kusukawa J, Irie K, Morimatsu M, Koyanagi S, Kameyama T. Dentigerous cyst associated with a deciduous tooth. A case report. *Oral Surg Oral Med Oral Pathol*. 1992;73:415-418.
11. Motokawa W, Braham RL, Morris ME, Tanaka M. Surgical exposure and orthodontic alignment of an unerupted primary maxillary second molar impacted by an odontoma and a dentigerous cyst: a case report. *Quintessence Int*. 1990;21:159-162.
12. Kishimoto H, Harada T, Yoshimura Y. Follicular cyst originated from deciduous molar in a 5-month-old infant: Report of a case. *Jpn J Oral Maxillofac Surg*. 1988;34:1394-1398 (in Japanese, abstract in English).
13. Rosenlicht J, Murphy JB, Maloney PL. Fetal alcohol syndrome. *Oral Surg Oral Med Oral Pathol*. 1979;47:8-10.
14. Yamanaka T, Sawakuma Y, Wakae H, Toyoshima S, Tomioka T. A case of follicular cyst probably due to a deciduous tooth. *Jpn J Oral Maxillofac Surg*. 1977;23:370-374 (in Japanese, abstract in English).
15. Neville BW, Damm DD, Chi AC. *Oral and Maxillofacial Pathology*. 4th Ed. St Louis: Saunders; 2015. p. 632-635.
16. Barnes L. *Surgical Pathology of the Head and Neck*, 3rd Ed. New York: Informa healthcare; 2009. p. 1201-1208.