

Infected Dentigerous Cyst Associated with Mesiodens: A Surprise for Maxillary Swellings

¹C Supreetha, ²SV Sreelatha, ³Arvind Karikal

ABSTRACT

Introduction: Dentigerous cysts are most commonly associated with unerupted third molars. Their association with a mesiodens is rare. Large swelling in the anterior maxillary region associated with severe destruction of bone is an unusual presentation of a dentigerous cyst when associated with an impacted tooth, especially a mesiodens.

Case presentation: This paper presents a case of a longstanding infected dentigerous cyst, associated with a mesiodens in the anterior maxillary region of a 32-year-old male patient. Radiographs revealed a large radiolucent lesion with extensive bone loss, thus implying the need for early diagnosis and intervention. Cone beam computed tomography (CBCT) revealed an associated mesiodens.

Clinical significance: Maxillary swelling arising as a result of an impacted mesiodens that clinically gave an impression of a space infection or a periapical cyst.

Treatment and prognosis: Enucleation of the cyst was done followed by the extraction of the mesiodens. Postoperative phase was uneventful and patient responded well to the treatment. Patient was advised for regular follow-ups.

Conclusion: Dentigerous cyst associated with the mesiodens is a rare entity. Complications, such as delayed or ectopic eruption or swelling in the area of the central incisors should alert the clinician to the possibility of a mesiodens. The clinician should obtain accurate radiograph to avoid missing out on the mesiodens. Early diagnosis of a mesiodens minimizes the treatment required and prevents development of associated problems.

Keywords: Anterior maxilla, Infected dentigerous cyst, Mesiodens, Supernumerary teeth.

How to cite this article: Supreetha C, Sreelatha SV, Karikal A. Infected Dentigerous Cyst Associated with Mesiodens: A Surprise for Maxillary Swellings. *Oral Maxillofac Pathol J* 2017;8(1):47-51.

Source of support: Nil

Conflict of interest: None

¹Postgraduate Student, ²Professor, ³Reader

^{1,2}Department of Oral Pathology and Microbiology, A B Shetty Memorial Institute of Dental Sciences, NITTE University Mangaluru, Karnataka India

³Department of Oral and Maxillofacial Surgery, A B Shetty Memorial Institute of Dental Sciences, NITTE University Mangaluru, Karnataka India

Corresponding Author: SV Sreelatha, Professor, Department of Oral Pathology and Microbiology, A B Shetty Memorial Institute of Dental Sciences, NITTE University, Mangaluru, Karnataka India

INTRODUCTION

A dentigerous cyst encloses the crown of an unerupted tooth by expansion of its follicle, and is attached to its neck.¹ Dentigerous cysts or follicular cysts, though the latter term is no longer used by many, progress by accumulation of fluids between the reduced enamel epithelium and the crown, with consequent expansion of the tooth follicle. They rank second in the list of most commonly occurring odontogenic cysts followed by radicular cysts.²

The age distribution of dentigerous cyst is seen between the first four decades of life. The highest frequency has been observed in the second and third decades. A substantial majority involved the mandibular third molar followed by the mandibular premolars and the maxillary third molars in order of frequency of involvement. This cyst occasionally involves the supernumerary teeth.²

A supernumerary tooth is an additional entity to the normal series of 10 deciduous and 16 permanent teeth in each quadrant and can be seen in all quadrants of the jaw.³⁻⁵ Mesiodens is one such supernumerary tooth that is present between the upper central incisors, hence its name. The incidence of the mesiodens involved by dentigerous cyst has been reported to be 0.3 to 0.8% in the primary dentition and 0.1 to 3.8% in the permanent dentition.^{6,7}

This paper presents a case of a dentigerous cyst associated with a mesiodens in the anterior maxillary region of a 32-year-old male patient.

CASE REPORT

A 32-year-old male patient reported with a chief complaint of swelling in the upper left side of the face since 3 years. Patient gave a history of a trauma to the mid-face 5 to 6 years back, but the swelling was present prior to the trauma. The swelling was observed by the patient to be with periods of regression following a pus discharge. The patient had no contributory medical history during his reporting of the problem.

Extraoral examination revealed a localized swelling in the left side of the face, lateral to the nose. Intraoral clinical examination revealed a soft labial and palatal swelling measuring 3.5 × 4.5 cm extending posteriorly. Palatally,



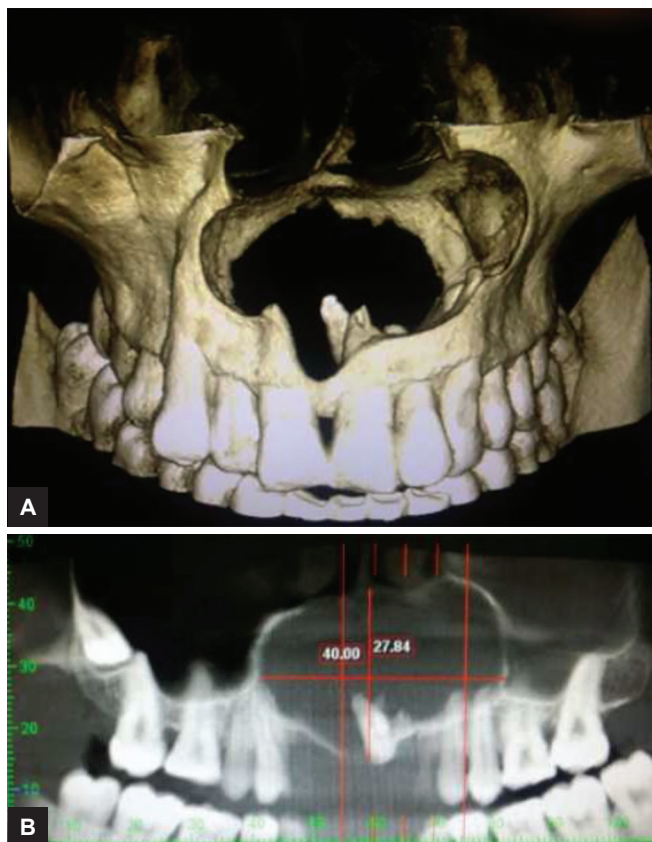
Fig. 1: Palatal swelling seen extending between 14 and 25

the swelling was seen extending between upper right first premolar and upper left second premolar. A presence of sinus tract was noted in relation to 11. The teeth 11, 14, 21, 23, and 24, however, did not respond to pulp vitality testing (Fig. 1). With the above clinical findings, radicular cyst was considered as a clinical differential diagnosis.

The panoramic radiograph showed presence of a well-defined radiolucent lesion with sclerotic borders. The extent of the lesion was from upper right second premolar to upper left first molar. Occlusal radiograph revealed the mesiodens placed palatally behind the upper left central incisor, which was not well appreciated in the panoramic radiograph. Cone beam computed tomography (CBCT) was also done which revealed an impacted mesiodens with respect to 11 and 21 and a radiolucency of 4×2.7 cm arising from the mesiodens extending from 14 to 24. Impacted 28 was also noted. External and external tooth resorption was seen in relation to 22. Three-dimensional reconstructions showed destruction of the labial plate, palatal plate, and floor of the nasal bone. A radiographic diagnosis of dentigerous cyst was given (Figs 2A and B).

The lesion was completely enucleated with the mesiodens under local anesthesia. The specimen received was fixed in formalin solution. Examination of the gross specimen revealed a hard tissue resembling a conical-shaped tooth, mesiodens, and cyst lumen attached to the neck of the tooth, measuring $2.5 \times 3.4 \times 0.7$ cm in size (Fig. 3).

Histopathological examination of the soft-tissue specimens revealed a nonkeratinized cystic epithelial lining, 2 to 4 cell layers thick made up of cuboidal cells, representing the reduced enamel epithelium and a fibrous connective tissue wall with a moderate chronic inflammatory component. Histopathologically, the diagnosis of infected dentigerous cyst was reported (Fig. 4).



Figs 2A and B: (A) Three-dimensional reconstructions showed destruction of the labial plate, palatal plate, and floor of the nasal bone; and (B) demonstrates a well-defined radiolucent lesion with the sclerotic borders arising from the mesiodens

Correlating the clinical, radiological, and histopathological findings, the final diagnosis of infected dentigerous cyst associated with mesiodens was given.

Cyst enucleation with the extraction of the mesiodens was performed under local anesthesia (Fig. 5) and an acrylic palatal plate was given in this case. Postoperative phase was uneventful and patient responded well to the treatment. Patient was advised for regular follow-ups.

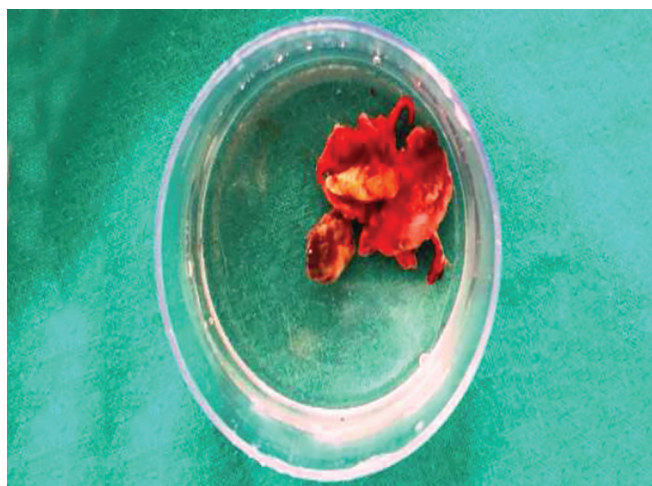


Fig. 3: Gross picture showing cystic lesion associated with the mesiodens

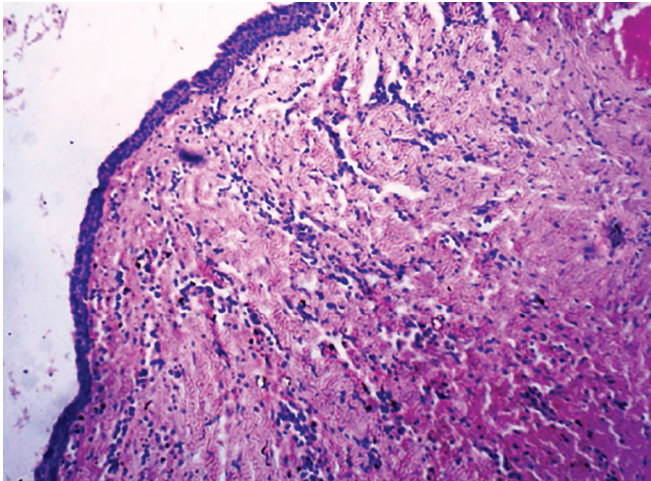


Fig. 4: Histopathological examination revealed the cyst lined by a stratified squamous nonkeratinized epithelium with a fibrous connective tissue wall. Hemotoxylin and eosin (H&E) staining; 4×

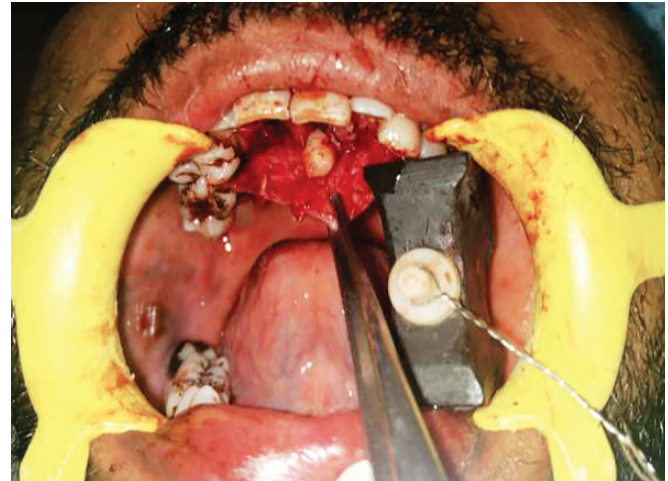


Fig. 5: Intra-operative image showing palatally placed mesiodens

DISCUSSION

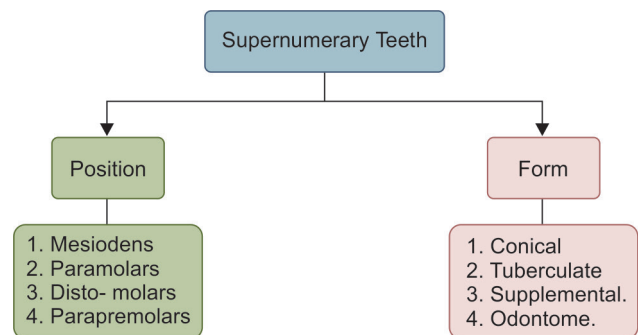
Dentigerous cysts are the second most common odontogenic cysts after radicular cysts and are most commonly seen associated with third molars and maxillary canines,⁷ rarely involving the supernumerary teeth. Supernumerary teeth are associated with many complications that include failure of the teeth to erupt, malalignment of the teeth, a midline diastema, formation of cyst with significant destruction of bone with resorption of roots of adjacent teeth.⁸⁻¹²

Dentigerous cysts may grow to a subsequent large size before they are diagnosed. Most of them are diagnosed on routine dental examination, when the patient complains of a missing or a tooth that has failed to erupt. Otherwise a slowly enlarging swelling or slight pain associated with the swelling brings the patient to the dentist, which leads to these cysts being discovered. Dentigerous cysts may occasionally be painful, especially when they are infected. Although the patients may give a history of slowly enlarging swelling, Steward has shown radiologically that lesions 4 to 5 cm in diameter may develop in 3 to 4 years.¹

By definition, a supernumerary tooth is one that is additional to the normal series and can be found in almost any region of the dental arch.¹³ 0.15 and 1.9% is the reported prevalence of supernumerary teeth in the general population, with males more commonly affected than females.¹⁴

Supernumerary teeth can be classified based on their form and position (Flow Chart 1).¹⁵

Mesiodens is the supernumerary tooth that occurs between the maxillary central incisors. They can be present individually or occur as multiples, in which case they are termed as mesiodentes. Studies have reported



Flow Chart 1: Classification of supernumerary teeth based on position and form.

that majority of the supernumerary teeth occur in the maxilla (80–90%).¹⁶

The etiology for the occurrence of mesiodens remains unclear, however, few theories have been suggested. The theory which was known as phylogenetic reversion (atavism) postulated that mesiodentes represented a phylogenetic relic of extinct ancestors who had three central incisors. This theory has now been largely discarded by embryologists. A second theory known as dichotomy suggests that the tooth bud is split to create two equal or unequal sections resulting in formation of two equal-sized teeth or one normal and one dysmorphic tooth, one of which is the mesiodens.¹⁷ Supporters of this theory believe that dichotomy represents complete gemination, which also occurs frequently in the anterior maxilla. The third theory, most widely supported, involves the hyperactivity of the dental lamina. According to this theory, remnants of the dental lamina or palatal offshoots of active dental lamina are induced to develop into an extra tooth bud, which results in a supernumerary tooth. Genetics is also thought to contribute to the development of mesiodentes, and such teeth have been diagnosed in twins, siblings, and sequential generations of a single family.¹⁶

Scolozzi et al¹⁸ reported an unusual case of a large dentigerous cyst associated with an impacted mesiodens, resulting in a slow-growing swelling in the upper lip. Khan et al¹⁹ also described an upper lip swelling caused by a large dentigerous cyst associated with mesiodens. Vosough et al²⁰ reported a painless swelling in the upper jaw caused by a dentigerous cyst associated with an impacted mesiodens. Dinkar et al described an unusually early presentation of multiple mesiodens with associated dentigerous cyst.²¹ Here in our case, the patient presented with a localized facial swelling along with a palatal swelling.

A number of lesions come to mind for the clinical differential diagnosis when swellings in the face and palate are considered. Space infections, cystic lesions, and lumps of neoplastic etiology are more likely to fall into this zone. The swelling with pus discharge and the acute onset of the symptoms pointed toward an infection. Here in our case differential diagnosis of dentigerous cyst, nasopalatine cyst, and periapical cyst was considered. Dentigerous cysts around supernumerary teeth account for 5% of all dentigerous cysts, most developing around a mesiodens in the anterior maxilla.⁶ The nasopalatine duct cyst falls in the group of nonodontogenic cysts which are not commonly associated with nonvital teeth.^{1,20} In this patient, history of trauma, and absence of response to the pulp vitality directed towards the diagnosis of periapical cyst. Due to the presence of impacted mesiodens and a cyst associated with it, ruled out periapical cyst. However, most radicular/periapical cysts appear as round or pear-shaped, unilocular, lucent lesions in the periapical region, and the associated tooth usually has a deep restoration or large carious lesion radiographically.⁸ In our case, another differential diagnosis of unicystic ameloblastoma could also be considered due to the destructive nature of the lesion, as it had eroded the buccal and the palatal bone. Unicystic ameloblastoma occurs in people under the age of 30 years, has equal gender predilection, and is most commonly associated with the crown of an unerupted third molar in the mandible.¹⁹

Cyst enucleation with the extraction of the supernumerary teeth and the restoration of the teeth affected is the treatment of choice, with regular recall check-up. As suggested by Scolozzi et al,¹⁸ large cysts can be treated by enucleation complemented by bone grafting.

CONCLUSION

Dentigerous cyst associated with mesiodens is a rare entity. Complications, such as delayed or ectopic eruption or swelling in the area of the central incisors should alert the clinician to the possibility of a mesiodens. The clinician should obtain accurate radiographs, including

panoramic, periapical, and occlusal views in order to avoid missing out on the mesiodens. Early diagnosis of a mesiodens minimizes the treatment required and prevents development of associated problems.¹⁶

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

AUTHORS' CONTRIBUTIONS

Arvind Karikal excised the tumor and C Supreetha, SV Sreelatha have contributed in analyzing, reading, writing, and researching this article. C Supreetha has done the research for the article guided by SV Sreelatha.

REFERENCES

1. Shear, M.; Speight, P. Cysts of the oral and maxillofacial regions. 4th ed. Blackwell; 2007. p. 59-75.
2. Neville, BW.; Damm, DD.; Allen, CM.; Bouquot, JE. Oral and maxillofacial pathology. 3rd ed. St. Louis: Saunders; 2008. p. 679-681.
3. Rajendran, R.; Sivapathasundharam, B. Shafer's textbook of oral pathology. 7th ed. Elsevier; 2012. p. 48-49.
4. Parolia A, Kundabala M, Dahal M, Mandakini M, Thomas MS. Management of supernumerary teeth. J Conserv Dent 2011 Jul;14(3):221-224.
5. Shah A, Gill DS, Tredwin CJ, Naini FB. Diagnosis and management of supernumerary teeth. Dent Update 2008 Oct;35(8):510-520.
6. Primosch RE. Anterior supernumerary teeth – assessment and surgical intervention in children. Pediatr Dent 1981 Jun;3(2):204-215.
7. Regezi, AJ.; Sciubba, JJ; Jordan, RCK. Oral pathology: clinical-pathologic correlations. 5th ed. St. Louis: Saunders; 2008. p. 242-244.
8. Kamboj M, Shreedhar B, Srivastava G, Verma D. Dentigerous cyst associated with mesiodens: a symbiotic existence. J Oral Health Comm Dent 2014 May;8(2):119-121.
9. Awang MN, Siar CH. Dentigerous cyst due to mesiodens: report of two cases. J Ir Dent Assoc 1989;35(3):117-118.
10. John T, Guna Shekhar M, Koshy M. Dentigerous cyst associated with supernumerary teeth: a report of three cases. J Clin Diagn Res 2010;4:2601-2606.
11. Stefane EC. Supernumerary upper central incisors. Dent Cosmos 1931;73(10):976-980.
12. Kumar NM, Ramadevi S, Vanaki SS, Puranik RS. Dentigerous cyst occurring in maxilla associated with supernumerary tooth showing cholesterol clefts – a case report. Int J Dental Clin 2010 Jun;2(2):39-42.
13. Garvey MT, Barry HJ, Blake M. Supernumerary teeth – an overview of classification, diagnosis and management. J Can Dent Assoc 1999 Dec;65(11):612-616.
14. Van Buggenhout G, Bailleul-Forestier I. Mesiodens. Eur J Med Genet 2008 Mar-Apr;51(2):178-181.

15. Arathi R, Ashwini R. Supernumerary teeth: a case report. *J Indian Soc Pedod Prev Dent* 2005 Jun;23(2):103-105.
16. Russell KA, Folwarczna MA. Mesiodens – diagnosis and management of a common supernumerary tooth. *J Can Dent Assoc* 2003 Jun;69(6):362-366.
17. Meighani G, Pakdaman A. Diagnosis and management of supernumerary (mesiodens): a review of the literature. *J Dent* 2010 Winter;7(1):41-49.
18. Scolozzi P, Lombardi T, Richter M. Upper lip swelling caused by a large dentigerous cyst. *Eur Arch Otorhinolaryngol* 2005 Mar;262(3):246-249.
19. Khan MH, Alam MT, Haque S, Khan SH, Fatema CN, Tahsin T, Choudhury AR. Upper lip swelling caused by a large dentigerous cyst with mesiodens. *Mymensingh Med J* 2008 Jul;17(Suppl 2):S100-S103.
20. Vosough Hosseini S, Moradzadeh M, Lotfi M, Aghbali AA, Fattahi S. Dentigerous cyst associated with a mesiodens: a case report. *J Dent Res Dent Clin Dent Prospects* 2011 Spring; 5(2):76-78.
21. Kumar M, Umashankar DN, Nanda Kumar, Radhika BM, Sudhakar R. Inflammatory variant of dentigerous cyst in maxillary sinus – a case report. *Int J Oral Maxillofac Pathol* 2010 Dec;1:17-19.