

Dentigerous cyst associated with a supernumerary tooth in the maxillary incisal region – a case report

*Erinosa OA, *Fadeyibi SO, **Gbotolorun MO, **Adeyemi MO

*Department of Oral and Maxillofacial Surgery, Lagos University Teaching Hospital, Idi-Araba, Lagos

**Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, College of Medicine, University of Lagos

Correspondence: Erinosa OA

Email: olufemierinosa@gmail.com

Abstract

Dentigerous cysts are defined as developmental odontogenic cysts, which surround the crown of an impacted tooth. It is caused by fluid accumulation between the reduced enamel epithelium and the enamel surface, resulting in a cyst with the tooth crown located within the lumen. This paper reviews the pathogenesis, clinical features and the behaviour of dentigerous cyst. It also presents a case of a slow growing, painless swelling in the maxillary incisal region of 3 years duration. Detailed clinical, radiological and histopathological investigations aided the definitive diagnosis of dentigerous cyst. The cystic mass was enucleated with the impacted tooth in situ under local anesthesia. The operative site healed uneventfully and a removable partial denture was fabricated to improve aesthetics, speech and masticatory function.

Key words: Odontogenic cysts, dentigerous cysts, supernumerary teeth, impaction

Introduction

Odontogenic cysts are pathological cavities lined by derivatives of the odontogenic epithelium, which are associated with the development of the dental apparatus⁽¹⁾. Dentigerous cysts are defined as developmental odontogenic cysts, which surround the crowns of impacted teeth, caused by fluid accumulation between the reduced enamel epithelium and the enamel surface, resulting in cysts with the tooth crown located within the lumen^(2, 3). Dentigerous cysts are generally reported to be the second most frequent type of odontogenic cysts accounting for 10- 20% of all jaw cysts⁽³⁻⁶⁾.

Dentigerous cysts are typically associated with the third molar teeth of the mandible and maxillary canines⁽¹⁻⁴⁾. They may also involve impacted supernumerary teeth in the anterior maxilla and rarely central incisors^(2, 7). Stafne,⁽⁸⁾ who was the first to describe an association between dentigerous cysts and supernumerary teeth, reported an incidence of 5.5% in 200 dentigerous cysts⁽⁴⁾. The most frequent age distribution is amongst individuals of the age group between 20 and 40 years, with male gender predominance⁽¹⁻⁴⁾.

The histopathologic appearance of the lining epithelium of dentigerous cyst is not specific⁽³⁾, the diagnosis therefore relies on the radiographic and surgical observation of the attachment of the cyst to the cemento-enamel junction of a partially or unerupted tooth⁽¹⁾. There are also several ways to manage this condition, ranging from marsupialization to surgical removal and extraction of the impacted tooth⁽³⁾.

Although enucleation of the cysts along with extraction of the tooth forms the mainstay of treatment and ensures complete removal, isolated lesions with preservation of the teeth and surrounding vital structures is desirable.

We present a rare case report of a dentigerous cyst, in the maxillary incisal region associated with an impacted supernumerary tooth in an adult male. This paper reviews the pathogenesis, clinical features and the behaviour of dentigerous cyst in an unusual location, and to our knowledge no case has been reported in Nigerian literature at this anatomical site.

Case Report

A 39 year old male presented at the Lagos University Teaching Hospital (LUTH) Lagos, with a slow growing, painless swelling in the anterior maxillary region of 3 years duration. He had no known systemic diseases, toxic habits, as well as allergies.

On extraoral examination, there was fullness of the upper lip and obliteration of the nasolabial fold on the left. Intra-orally, there was a well-defined oval swelling, extending from the distal aspect of 11 to the 23, measuring 3 x 2.5cm, with labio palatal expansion (**Figure 1**). The mucosal surface appeared smooth and showed no discolouration. The patient had full complement of teeth.



Figure 1. Intraoral view of the 39 year old male

On palpation, the swelling was fluctuant, not tender, with no differential warmth. 21 and 22 were discoloured with associated grade 3 mobility. The discoloured teeth tested negative to pulp vitality tests. On aspiration, a straw coloured fluid was obtained and sent for histopathological analysis. Clinical impressions of radicular, dentigerous and primordial cysts were made.

An orthopantomograph (OPG) revealed a radiolucent image about 4mm apical to the roots of the 21 and 22 surrounding an impacted supernumerary incisor tooth (**Figure 2**). A provisional diagnosis of dentigerous cyst was made, and nasolabial cyst was listed as a differential.

An enucleation of the mass with the impacted tooth was done under local anesthesia. 21 and 22 were extracted (**Figure 3**) and the resulting cavity was curretted and irrigated thoroughly with 0.9% Normal saline. Suturing was done with 4/0 silk sutures.



Figure 2. OPG of the 39 year old male



Figure 3. The cystic mass enucleated

Histopathology report of the specimen detailed a cystic cavity lined by stratified squamous epithelium of 2-3 cell layers thick, overlying a fibrous connective tissue within. Other cells such as chronic inflammatory cells, consisting of lymphocytes, histiocytes, and plasma cells were seen. Also seen were endothelial-lined vascular spaces. A diagnosis of dentigerous cyst was confirmed (**Figure 4**).

Figure 5 shows the intra-oral view of the patient 2 weeks after the operation. Regular post-operative follow-up reviews revealed a favourable clinical and radiological evolution of the procedure. After 1 month, a removable partial denture was delivered (**Figure 6**), which improved esthetics, speech and served as a space maintainer.

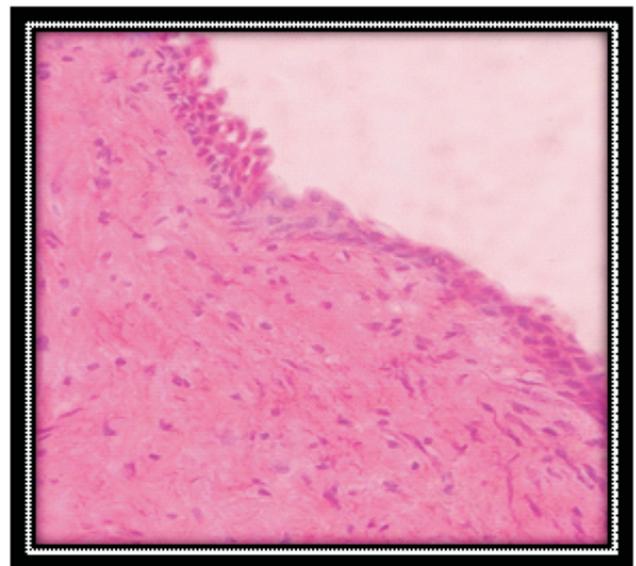


Figure 4. Histological appearance of the lesion



Figure 5. 2 weeks intra-oral view after the operation



Figure 6. 1 month post operatively after fabrication of upper removable partial dentures.

Dentigerous means containing teeth^(7,9). The incidence of dentigerous cyst is 20 to 24% of the entire epithelial-lined jaw cysts^(1,7).

A majority of these cysts has been reported to affect the mandibular third molar and the maxillary permanent canine, followed by the mandibular premolars, maxillary third molars and rarely the central incisors^(2,10).

In the past, various studies confirmed the rarity of these cases in the anterior maxillary region⁽¹¹⁾. According to Kalaskar et al⁽²⁾ the incidence of dentigerous cysts involving the maxillary central incisor was 1.5% as compared to 45.7% involving the mandibular third molar. A review of literature by Jiang et al⁽¹⁰⁾ using Medline, detailed the incidence of dentigerous cysts associated with anterior maxillary supernumerary teeth between 1988 and 2011 to be 16 cases. Also reported as unusual is the association between dentigerous cysts and supernumerary teeth^(4,9,10,12)

and several theories have been propounded for the occurrence⁽¹³⁾.

A retrospective study by Hurlen and Humerfelt⁽¹⁴⁾, detailed an incidence of 7% in supernumerary teeth which developed into dentigerous cysts. This was supported by Lin et al⁽¹⁵⁾. Although association of dentigerous cysts with supernumerary teeth still remains an unusual pathology^(4,9,10), dentigerous cysts should be an important differential diagnosis in swellings of the anterior maxillary region, especially when supernumerary teeth have been identified.

Dentigerous cysts most commonly occur in the first four decades of life⁽⁹⁾. They have consistently shown a male predilection as seen in this reported case, and a predominance in Caucasian and Asian races as compared to Africans⁽¹²⁾. Perhaps this has contributed to its rarity in our environment, and to our knowledge no case has been reported in Nigerian literature at this anatomical site.

Clinically, dentigerous cysts usually present as, solitary, slow growing, expansile, asymptomatic lesions⁽¹²⁾. Patients may complain of swelling along the palatal surface or on the upper lip if in the anterior maxilla, a missing or partially erupted tooth and pain in rare cases of superimposed infection^(9,10). Most authors have reported the presence of carious or discoloured deciduous teeth in relation to the development of dentigerous cysts^(2,11,16). This suggests that the periapical inflammatory exudate from the teeth might be one of the risk factors for the occurrence of dentigerous cysts^(2,17).

Panoramic and upper occlusal radiographs are simple and readily available methods⁽¹⁰⁾, as used in this case reported in this paper. However, the use of plain radiograph has the disadvantages of an inability to reflect the 3-dimensional structure of the lesion owing to a low resolution ratio, and different degrees of distortion or amplification⁽¹⁰⁾. Despite these, the aforementioned plain views remain the first line of diagnostic tools, after which a CT image can be obtained. Dentigerous cysts appear as a unilocular radiolucency with a well circumscribed sclerotic border and an embedded radio opacity of an unerupted tooth^(1,18). In this case report, the radio opaque tooth structure was attached to the radiolucency at the cemento enamel junction, and there was resorption of adjacent bone and roots of surrounding teeth.

Histologically, the lumen is usually lined by 2 to 4 cell layers of squamous non-keratinized epithelial cells^(1,13). The epithelium may be hyperplastic with the presence of hyaline bodies associated with inflammation⁽¹²⁾. The connective tissue, when inflamed may contain varying degrees of chronic inflammatory cell infiltration, and the cyst lining may contain ciliated and mucous secreting cells^(1,4,12). These features were also observed histologically in our reported case.

In the management of dentigerous cysts, early detection and surgical removal of the cysts are important to reduce morbidity^(12,13). Treatment of dentigerous cysts depends on the size, site, patient's age and degree of involvement of surrounding vital structures⁽¹⁹⁾. Treatment options can

include enucleation along with extraction of the associated supernumerary tooth as done in our case. Other options include marsupialization and decompression, when surrounding vital structures may be damaged by enucleation⁽²⁰⁾. In event of a large defect following surgical enucleation, a bone graft may be necessary⁽⁴⁾.

Complications associated with dentigerous cysts include pathological bone fracture particularly in large cysts, loss of the permanent tooth following management as a result of extensive local bone resorption and loss of tooth vitality which was experienced in our case, and bone deformation⁽⁹⁾. The lining epithelium of the cyst has also been reported to have pluripotential capacity, therefore, these lesions may progress to ameloblastoma, mucoepidermoid carcinoma and squamous cell carcinoma if improperly managed⁽¹²⁾.

Conclusion

Dentigerous cyst associated with a supernumerary tooth in the maxillary central incisal region is a rare entity. A detailed history, physical and radiographic examination is of importance, and will ultimately guide the definitive surgical treatment and follow-up. The result of our approach to the management of this case was the restoration of function in the oral apparatus and aesthetics in the facial profile.

Reference

1. Rohilla M, Marwah N, Tyagi R. Anterior Maxillary Dentigerous Cyst- A case report. *Int J Clin Pediatr Dent* 2009; 2:42-45.
2. Kalaskar RR, Tiku A, Damle SG. Dentigerous cysts of anterior maxilla in a young child: A case report. *J Indian Soc Pedod Prev Dent* 2007; 25: 187- 190.
3. Shafer WG, Hine MK, Levy BM, Rajendran R, Sivapathasundharam B (eds). *A textbook of Oral Pathology*. 5th ed. New Delhi: Reed Elsevier 2006; 358- 362.
4. Navarro BG, Salas EJ, Olmo IT, Muñoz AF, Escalona IJ, López-López JL. Maxillary dentigerous cyst and supernumerary tooth. Is it a frequent association? *Oral Health Dent Manag* 2014; 13: 127-131.
5. Ogunlewe MO, Odukoya O, Akinwande JA. Epithelial jaw cysts: analysis of 126 Nigerian cases. *Afr Dent J* 1996; 10:1-8.
6. Iyogun CA, Ochicha O, Sule AA, Adebola RA. Jaw Cysts in Kano; Northern Nigeria. *Int J Oral Maxillofac Pathol* 2013; 4:8-12.
7. Desai RS, Vanaki SS, Puranik RS, Tegginamani AS. Dentigerous cyst associated with central incisor teeth: A rare entity. *J Indian Soc Pedo Prev Dent* 2005; 49-50.
8. Stafne EC. Supernumerary upper central incisor. *Dental Cosmos*. 1931; 73: 976-980.
9. Baranwal HC, Verma PK, Dwivedi CD, Srivastava R. Dentigerous cyst associated with an impacted maxillary mesiodens. *Eur J Gen Dent* 2012; 1:50-53
10. Jiang Q, Xu G, Yang C, Yu C, He D, Zhang Z. Dentigerous cyst associated with impacted supernumerary teeth in the anterior maxilla. *Exp Ther Med* 2011; 2:805-809.
11. Mourshed F. A Roentgenographic study of dentigerous cyst: Incidence in a population sample. *Oral Surg Oral Med Oral Pathol* 1964; 18:54-61.
12. Mishra R, Tripathi AM, Rathore M. Dentigerous cyst associated with horizontally impacted mandibular second premolar. *Int J Clin Pediatr Dent* 2014; 7: 54-57.
13. Nair P, Khare P. Dentigerous cyst associated with impacted supernumerary maxillary anterior tooth. *J Orofac Research* 2013; 3: 280-283
14. Hurlen B, Humerfelt D. Characteristics of premaxillary hyperdontia. A radiographic study. *Acta Odontol Scand* 1985; 43: 75-81.
15. Lin HP, Wang YP, Chen HM, Cheng SJ, Sun A, Chiang CP. A clinicopathological study of 338 dentigerous cyst. *J Oral Pathol Med* 2013; 42: 462-467.
16. Wang CJ, Huang PH, Wang YL, et al. Dentigerous cyst over maxillary sinus: a case report and literature review. *Taiwan J Oral Maxillofac Surg* 2009; 20:116-124.
17. Benn A, Altini M. Dentigerous cysts of inflammatory origin. A clinicopathologic study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1996;81:203-209
18. Sharma A, Singh VP, Sharma S. Dentigerous cyst associated with unusual supernumerary tooth: a case report. *Asian J MedSci* 2011; 2:124- 127.
19. Motamedi MHK, Talesh KT. Management of extensive dentigerous cysts. *Br Dent J* 2005; 198:203-206
20. Ziccardi VB, Eggleston TE, Schnider RE. Using fenestration technique to treat a large dentigerous cyst. *J Am Dent Assoc* 1997;128:201-205