Recurrent Dentigerous Cyst with Malignant Transformation of Cyst Lining – A Case Report

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Dentigerous cyst is the most common of the developmental odontogenic cysts of the jaws. In addition to theirpotential for recurrence and attaining large size, dentigerous cysts are noteworthy for the occasional development ofameloblastoma and malignancy in an isolated segment of the cyst wall. This potential for neoplastic change fully justifies the need for a thorough histopathologic examination. Here, we report a case of recurrent dentigerous cyst in posterior mandible clinically and radiographically presenting as an ameloblastoma, which was confirmed on histopathological examination to be a dentigerous cyst with malignant transformationof the cyst lining.

Keywords: Dentigerous cyst, Ameloblastoma, Mural ameloblastoma, Developmental cyst

Odontogenic cysts arise from an aberration occuring at some stage of odontogenesis.[1] Dentigerous cysts are the most common of the developmental odontogenic cysts of the jaws.[2,3,4,5,6,7]They account for approximately 20 – 24% of all the epitheliumlined jaw cysts.[2, 3,6,7, 8,9] The WHO classification of epithclial cysts related to the odontogenic apparatus refers to dentigerous or follicular cyst as a 'developmental odontogenic cyst'.[10, 11] Dentigerous simply means containing teeth.[8]A dentigerous cyst can be defined as a cyst that encloses the crown of an unerupted tooth, expands the follicle and attaches to the cementoenamel junction of the unerupted tooth,[2, 3, 4, 5, 6, 11, 12, 13] originatingby accumulation of fluid either between the reduced enamel epithelium and enamel or in between the layers of the enamel organ.[2,5,7,8,14,15,16]

Dentigerous cysts most commonly occur in the 2nd and 3rd decades of life,[1,2,3,4,5,10,14,17] though they may also be found in children. Their occurrence has a definite male predilection.[1,2,3,4,5,9,14,18] Dentigerous cysts are found most frequently around the crown of impacted mandibular third molars followed, in order of frequency, by the maxillary canines, maxillary third molars and rarely, supernumerary teeth and central incisors.[1,2,3,6,8,9,16,19,20]

These cysts are often asymptomatic unless there is an acute inflammatory exacerbation, and therefore, these lesions are usually diagnosed during routine radiographic examination of a missing tooth.[1,2,6,9,10,13,14,15,18] Swelling, tooth displacement, tooth mobility, root resorption and tooth sensitivity may be present if the cyst reaches a size larger than 2 cm in diameter.[2] They may expand to a substantial size and may cause retention of any involved teeth, deformation of surrounding alveolar bone and expansion of the cortical bone.[7,10,15] However, destruction of the bone is rare.[10] Most dentigerous cysts are solitary, butmultiple cysts may befound in association with syndromes such as cleidocranial dysplasia, mucopolysaccharidosis, basal cell nevus syndrome and Maroteaux Lamy syndrome.[1,9,17,19]

While a normal follicular space is 3 to 4 mm, a dentigerous cyst can be suspected when the space is more than 5mm.[1, 9,10, 13, 15, 20] The cyst appears radiographically as a well-demarcated radiolucent lesion attached at an acute angle to the cervical area of an unerupted tooth. The border of the lesion may be radiopaque.[1,2,5,6,9,10,12,13,15,17,19]

Microscopically, it reveals a fluidfilled cyst lined by a thin, keratinized epitheliumconsisting of 2 to 6 layers of cuboidal epithelial cellsand flat epithelium –

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connectivetissue interface. The inflammatory cyst exhibits mononuclear cells in the cystic wall, with a possibility of squamous metaplasia of the epithelium.[2,9,10,12,14] The content of the cyst lumen is usually a thin, watery yellow fluid, but may be blood tinged.[21]

The differential diagnosis of dentigerous cyst includes radicular cyst, inflammatory paradental cyst, odontogenic keratocyst, idiopathic bone cavity and odontogenic tumours such as the unicystic ameloblastoma, mural ameloblastoma, ameloblastic fibroma, Pindborg tumour, odontogenic fibroma, adenomatoid odontogenic tumour, odontogenic fibromyxoma, odontoma andcentral giantcell granuloma.[2,7,9,15,20]

The choice of the therapeutic approach should be based on the size and location of the cyst, the patient's age, affected dentition and relationship with surrounding vital structures.[8,20] The treatment options are enucleation with or without removal of the involved tooth or marsupialization.[2,7,8,12,13,14,15,18,19,20]

Complications associated with dentigerous cysts include pathologic bone fracture, loss of the permanent tooth, bone deformation[2,3,6] or transformation of cyst lining into an odontogenic tumour (e.g., ameloblastoma or mucoepidermoid carcinoma) or a malignancy (e.g., oral squamous cell carcinoma).[2,3]

Ameloblastoma is a benign, locally invasive tumor of epithelial origin known for its high recurrence rate.[22-24] Most cases of mural ameloblastoma occur in the third to fifth decade of life,[21-24] with no definite sex predilection.[21] The most common location is the posterior mandible, associated with impacted third molars.[21-24] The clinical presentation is often a slowly enlarging asymptomatic facial swelling.[21,24] The radiographic appearance varies from soap bubble loculations to an expansile unilocular or multilocular pattern with discrete margins.[??]

Here, we present a case of a recurrent dentigerous cyst clinically and radiographically simulating an ameloblastoma, which was confirmed histopathologically as a dentigerous cyst with malignant transformation of the cyst lining.

CASE REPORT

A 35 year old male patient reported to the Oral Medicine and Radiology Department of Ahmedabad Dental College and Hospital with the chief complaint of pain and swelling on right side of face since last 1 month. [Fig. 1] The swelling slowly grew to the present size within last monthassociated with pain on mastication in same region.

Patient was relatively asymptomatic before 9 years. Then he had pain and swelling in lower right posterior region. It was diagnosed clinically and radiographically as a dentigerous cyst associated with a mesioangular impacted 48. The cyst along with 46, 47 and impacted 48was surgically removed.Clinical diagnosis of dentigerous cyst was confirmed by the histopathological report. There was no other significant past medical or dental history. Patient had habit of tobacco placement in the form of quid combined with slaked lime and betel nut in the lower labial vestibule adjacent to 31, 32, 41& 42 about 3 – 4 times a day for 5 years. Patient had ceased the habit since last 2 years.

Extraoral examination revealed an ill – defined ovoid swelling sized 5 x 4 cm extending anteroposteriorly from corner of mouth upto lobule of ear and superoinferiorly from ala canthus line upto lower border of mandible. [Fig. 1] The swelling was firm, tender, non-warm andnon-pulsatilewith no anaesthesia or paresthesia of the overlying skin. There was no regional lymphadenopathy. Patient's mouth opening was adequate (34 mm).

Intraoral examination revealed that all teeth were present except 46, 47 & 48. [Fig. 2] A single, ill defined, lobulated, firm, tender, non-warm, non-pulsatile swelling sized 3x2 cm extended antero-posteriorly from 45 region upto anterior border of ramus and beyond and supero-inferiorly from coronoid process upto angle of mandible with expansion of buccal and lingual cortices and obliteration of buccal vestibule adjacent to 46, 47

and 48 region. There was redness and indentation of cusp tips of 16 & 17 on the superior surface of the swelling withfluctuancy in retromolar region and fixity to underlying structures.

On pulp vitality testing, 45 was non vital. Radiographicexamination included mandibular right cross sectional occlusal, OPG, PA Mandible and MDCT of facial bones. The mandibular right cross sectional occlusal radiograph confirmed expansion of buccal & lingual cortical plates. [Fig. 3] OPG revealed two radiolucencies in right mandibular body ramus region. The smaller one was unilocular extending antero-posteriorly from distal aspect of 45 upto 47 region and superoinferiorly from edentulous ridge of 46 region upto lower mandibular border. Posterior to the first, second larger radiolucency was well demarcated and multilocular (soap bubble appearance) with thinning and expansion of anterior and posterior borders of ramus. [Fig. 4] PA Mandible confirmed the above findings with 4 septa visible within the radiolucency giving it a multilocular appearance. It was sparing the right mandibular condyle. The MDCT of facial bones revealed a 50 x 35 mm expansile lytic lesion in right mandibular body-ramus region upto coronoid process with thinning as well as destruction of adjacent cortex. [Fig. 5] There was no evidence of significant lymphadenopathy or infiltration of adjacent structures.Based on clinical and radiographic examination, a provisional diagnosis of ameloblastoma was made. Differential diagnosis included dentigerous cyst and odontogenic keratocyst.

Fine needle aspiration produced a blood tinged fluid, the cytology report of which was unproductive. The incisional biopsy report revealed the swelling to be a dentigerous cyst.

As the lesion was extensive and involved a major portion of right half of mandible, hemimandibulectomy from 43 region was done under general anaesthesia. Level I and II nodes were also removed and sent for histopathological examination along with the excised cyst and portion of mandible.

The histopathological report of excised specimen confirmed the diagnosis of dentigerous cyst with a malignant transformation of the cyst lining. The cyst lining consisted of 2 – 4layers of flattened non keratinizing cells and the cystic cavity was filled with necrotic material. [Fig. 6] At some places luminal thickening with dysplastic changes like hyperchroma-tism, altered nucleo cytoplasmic ratio were also seen. The lymph nodes and salivary glandsshowed no malignant changes.

Post operatively, the patient was well with satisfactory healing of the surgery site.He is currently under regular follow up for post-operative prosthetic rehabilitation.

DISCUSSION

The present case was that of adentigerous cyst in a 35 year old male patient. The age and sex of the patient was in accordance with literature.[1,2,3,4,5,9,10,14,17,18] There was a history of occurrence of the same before 9 years in association with an impacted 48 which was confirmed histopathologically.

The site of swelling was in mandibular molar ramus region which is the most common site reported for occurrence of dentigerous cyst.[1,2,3,6,8,9,16,19,20] The swelling had a lobulated surface with fixity to underlying tissues with palpable expansion of cortical plates and widespread involvement of the mandible without regional lymphadenopathy. These clinical features are in accordance withdentigerous cyst.[7,10,15]

OPG, PA Mandible and MDCT findings revealed that the lesion was a multilocular one with thinning and destruction of cortical plates and anterior and posterior borders of ramus. These findings were in accordance with the provisional diagnosis of ameloblastoma.[22]The fact that dentigerous cyst has reported a high recurrence rate and transformation into an ameloblastoma supported by the radiographic features led us to place a diagnosis of amcloblastoma rather than a dentigerous cyst.[1,10,11]

Histopathological report of the incisional biopsy confirmed the lesion to be a dentigerous cyst. The

patient was treated with distal hemimandibulectomy because of recurrenceand the extensive bone destruction with very thin remaining bony cortices as revealed by MDCT. The histopathological examination of the excised specimen confirmed the diagnosis of dentigerous cyst with malignant transformation of the cyst lining, which justified the decision of treating the lesion radically rather than conservatively.[5,8,18]

CONCLUSION

"If knowledge is wealth, experience is wisdom". This case report provides an example of possible aggressive behaviour of a dentigerous cyst which may be mistaken clinically and even radiographically to be an ameloblastoma. Hence, a thorough history combined with clinical and radiographical examination of any aggressive lesion ismandatory. Also any surgical intervention should strictly be preceded by proper laboratory investigations and histopathological confirmation as malignant transformation is a possible complication.

References

1. Puneet Batra, Ajoy Roychoudhury, Prahlad Balakrishan, Hari Parkash. Bilateral dentigerous cyst associated with polymorphism in chromosome 1qh+. The Journal of Clinical Pediatric Dentistry, 2004; 28(2): 177 – 81.
2. Kalaskar R., Tiku A., Damle G. Dentigerous cysts of anterior maxilla in a young child: A Case Report. ISPPD, 2007: 187 – 90.
3. Naveen Kumar. M, Rama Devi. S, Shreenivas Vanaki. S, Puranik. R.S. Dentigerous Cyst Occurring in Maxilla Associated with Supernumerary Tooth Showing Cholesterol Clefts - A Case Report. International Journal of Dental Clinics, 2010; 2(2): 39 – 42.
4. Srinivasa Prasad T, Sujatha G, Thanvir Mohammad Niazi, Rajesh P. Dentigerous cyst associated with an Ectopic Third Molar in the Maxillary Sinus: A Rare Entity. Indian J Dent Res, 2007; 18(3): 141 – 3.
5. S.J.J. McCrea, Makdissi. A Comparison of natural bone regeneration with that produced by the Xenograft Bio-Oss in the management of an unusual case of adjacent Dentigerous Cysts. Oral Surgery, 2009; 2: 136 – 43.
6. Juliana Yuri Kawamura, Roberto Pereira de Magalhaes, Suzana Sousa, Marina Magalhaes. Management of a large dentigerous cyst occurring in a six-year old boy. The Journal of Clinical Pediatric Dentistry, 2004; 28(4): 355 – 8.
7. Eduardo Sannomiya, Marcelo Nogueira, Marli Diniz, Francisco Pacca, Gisele Dalben. Trauma induced Dentigerous Cyst Involving the Anterior Maxilla. Journal of Dentistry for Childrenm 2007; 74(2): 161 – 4.
8. Shivaprakash P, Rizwanulla T, Baweja D, Noorani HH. Save-a-tooth: Conservative surgical management of dentigerous cyst. J Indian Soc Pedod Prevent, 2009; 27(1): 52 – 7.
9. Sergio Cury, Maria Cury, Flavia Pontes, Helder Pontes, Camila Rodini. Bilateral Dentigerous Cyst in a Nonsyndromic Patient: Case Report and Literature Review. Journal of Dentistry for Children, 2009; 76(1): 92 – 6.
10. Maria Homem, Wilma Simoes, Maria Deboni, Israel Chilvarquer, Andreia Traina. Dentigerous cyst associated with an upper permanent central incisor: case report and literature review. The Journal of Clinical Pediatric Dentistry, 2002; 26(2): 187 – 92.
11. Haruaki Hayasaki, Mayuko Ishibashi, Seiji Nakamura, Satoshi Fukumoto, Kazuaki Nonaka. Dentigerous Cyst in Primary Dentition: Case report of a 4-year-old girl. Pediatric dentistry, 2009; 3(1): 294 – 7.
12. Kuo- Ting Sun, Michael Y. Chen, Hsien Chiang, Hung Tsai. Treatment of Large Jaw Bone Cysts in Children. Journal of Dentistry for Children, 2009; 76(3): 217 – 22.
13. Passi S, Gauba K, Agnihotri A, Sharma R. Dentigerous cyst in primary dentition: a case report. J Indian Soc Pedod Prevent Dent, 2008: 168 – 70.
14. Alkhatib, D. J. Manton. Case report: Preservation of teeth involved with an odontogenic cyst. European Archives of Paediatric Dentistry, 2010; 11(3): 146 – 8.
15. J. Berden, G. Koch, C. Ullbro. Case Series: Treatment of large dentigerous cysts in children. European Archives of Paediatric Dentistry, 2010; 11(3): 140 – 5.
16. Dentigerous cyst associated with trauma to a primary incisor: a case report. Constance M. Killian, Paul H. Leventhal, James L Tamaroff. Quintessence International, 1992: 23(10): 683 – 6.
17. Metachronous Bilateral Dentigerous Cysts Associated With Permanent First Molars. Eduardo Fregnani, Danyel Perez, Paulo Carvalho, Fabio Alves. Journal of Dentistry for Children, 2008; 75(2): 197 – 200.
18. Koca Huseyin, Alpoz Esin, Kazanc Aycan. Outcome of Dentigerous Cysts Treated with Marsupialization. The Journal of Clinical Pediatric Dentistry, 2009; 34(2): 165 – 8.
19. N. Shah, H. Thuau, T. Beale. Spontaneous regression of bilateral dentigerous cysts associated with impacted mandibular third molars. British Dental Journal, 2002; 192(2): 75 – 6.
20. Juliana Gondim, Jose Jeova Siebra Moreira Neto, Renato Nogueira, Elisa Giro. Conservative management of a

dentigerous cyst secondary to primary tooth trauma - Case Report. Dental Traumatology, 2008; 24: 676 – 9.

21. R. Rajendran, B. Sivapathasundaram. Shafer's Textbook of Oral Pathology, 6th Edition. Elsevier, 2009.

22. Chen YN, Nambiar P. Radiological Features of Different Histopathological Variants of Ameloblastomas. Malaysian Dental Journal 2008; 29(1): 14-19.

23. Akinola Ladipo Ladeinde, Mobolanle Olugbemiga Ogunlewe, Babatunde Olamide Bamgbose, Wasiu Lanre Adeyemo, Oluseyi Folake Ajayi, Godwin Toyin Arotiba, Jelili Adisa Akinwande. Ameloblastoma: Analysis of 207 cases in a Nigerian teaching hospital. Quintessence International 2006; 37(1): 69 – 74.

24. Martin Greenberg, Michael Glick, Jonathan Ship. Burket's Oral Medicine, 11th Edition. BC Decker Inc, 2008.

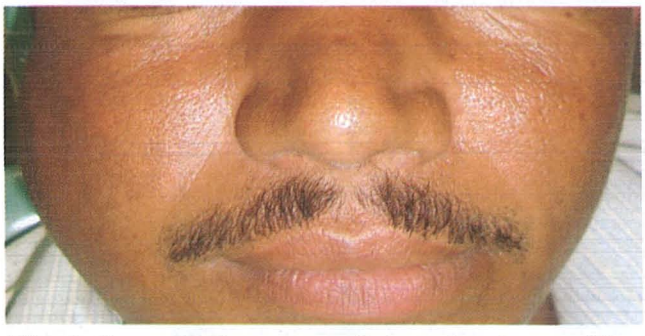


Fig. 1: Preoperative extraoral view showing swelling on right side of face



Fig. 2: Intraoral view showing extent of lesion superiorly and posteriorly



Fig. 3: Occlusal radiograph showing expansion of cortices

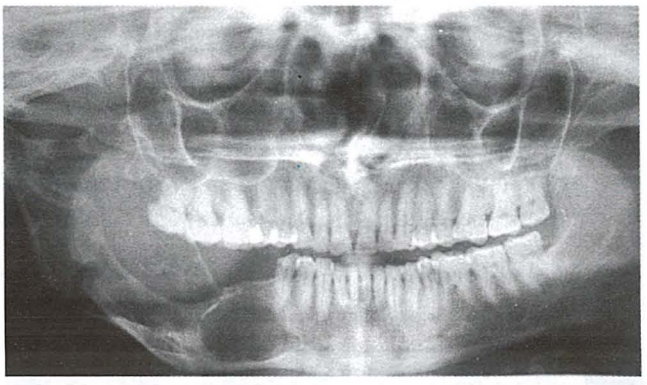


Fig. 4: OPG showing extent of the multilocular radiolucency in right molar ramus region of mandible

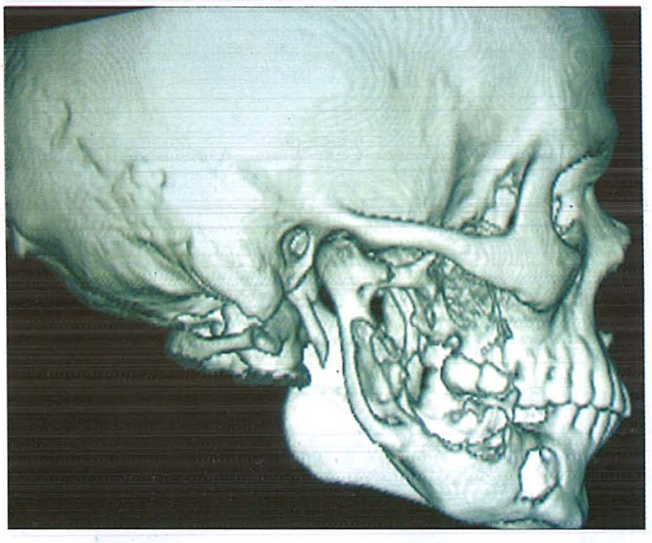


Fig. 5: MDCT 3D reconstruction of skull showing the destructive lesion

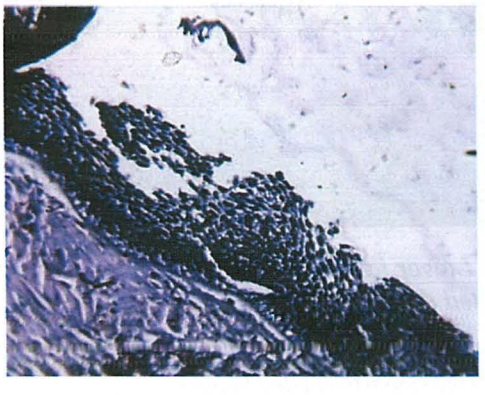


Fig. 6: Microphotograph at 40 X magnification of the section prepared from the excised specimen showing cyst lining characteristic of dentigerous cyst