

Marsupialization of odontogenic keratocyst using thermoform surgical splint in a pediatric patient, with 3-year follow-up

Srinivas Nallanchakrava, Mallela George Manoj Kumar, Dwitha Animireddy, Garikina Manasa

Department of Pediatrics and Preventive Dentistry, Panineeya Mahavidyalaya Institute of Dental Sciences and Research Centre, Hyderabad, Telangana, India

Abstract

Odontogenic keratocyst (OKC), also known as keratocystic odontogenic tumor, is categorized as one of the developmental epithelial cysts, which accounts 10%–20% of all cystic lesions in jaws. Considering its high recurrence rate, combative treatment modalities such as enucleation and jaw resection are suggested. This case report aims to emphasize clinical, radiological and histological overview of OKC with conservative approach marsupialization using thermoform surgical splint in a 9-year-old female patient with no recurrence at 3-year follow-up.

Keywords: Marsupialization, odontogenic keratocyst, thermoform splint

Address for correspondence: Dr. Srinivas Nallanchakrava, Department of Pediatrics and Preventive Dentistry, Panineeya Mahavidyalaya Institute of Dental Sciences and Research Centre, Kaloji Narayana Rao University of Health Sciences, Hyderabad - 500 060, Telangana, India.

E-mail: cnudent@gmail.com

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INTRODUCTION

Odontogenic keratocyst (OKC) that originates either from a dental lamina or primordial odontogenic epithelium^[1,2] is a benign tumor of the jaws, and the term was introduced by Philipsen in 1956.^[1,3] According to the World Health Organization's (WHO) 1992 classification, OKCs are typed under developmental (dysembryogenetic) cysts^[3] and were designated in 2005 as a keratocystic odontogenic tumor^[4,5] which is defined as "a benign uni or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium, and potential for aggressive, infiltrative behavior."^[5,6] In 2017, the fourth edition of the WHO/IARC Classification of Head and Neck Tumors were published where keratocystic odontogenic tumor is back into the cystic category as OKC.^[1,7] It is

frequently reported in the second to fourth decade of life and relatively less in the first decade.^[2] It has a multicentric growth pattern, infiltrating through cortical bone and extending into adjacent soft tissue.^[8] Conservative treatments are widely employed in children to avoid complications associated with radical operations.^[9] Marsupialization is one such conservative treatment, proved to be effective treating cysts among children and adolescents which secures adjacent vital anatomical structures such as inferior alveolar nerve, maxillary sinus and developing dentition.^[10]

CASE REPORT

A 9-year-old female patient reported to the Department of Paediatric and Preventive Dentistry with a complaint

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of pain and swelling in right lower back tooth region. On general examination, the patient was healthy. The patient had a history of pulp therapy in 84, 3 years back, and complained pain in the same region. Extraoral examination revealed facial asymmetry due to swelling noted on lower right side of face, with no visible pulsations or pus discharge [Figure 1a]. A single diffuse swelling, approximately 3 cm × 3 cm in size, extended anteriorly 1 cm behind the right corner of mouth to 3 cm posteriorly. Superiorly, extended 3 cm below the ala-tragal line and inferiorly to lower border of mandible. No paraesthesia was noted. Submandibular lymph nodes on right side were palpable, tender, mobile and firm in consistency. On intraoral examination, vestibular obliteration in right mandibular region from permanent lateral to the first molar with normal overlying mucosa was seen [Figure 1b]. Swelling was tender, nonfluctuant, bony hard in consistency. Other clinical findings included disto-proximal caries in 74 and deep occlusal caries in 36 [Figure 1b and c]. Panoramic radiography and cone beam computed tomography scan revealed a single homogenous well-defined radiolucency on the right body of mandible measuring approximately 2.5 cm × 3 cm in size, extending antero-posteriorly from distal aspect of 83 to distal side of 46 and expansion of the lesion resulted in displacement of permanent canine tooth bud downward and forward below incisors and premolars tooth buds downward and backward below permanent molar, 1 cm above the inferior border of mandible [Figure 1c]. The roots of deciduous first and second molar were resorbed. Inferior alveolar canal was not seen. Expansion of buccal and lingual cortical plates on the right side of mandible was observed [Figure 1d]. Based on clinical and radiological findings, the provisional diagnosis of radicular cyst was made. Histological examination was planned to differentiate from OKC, dentigerous cyst,

buccal bifurcation cyst and unicystic ameloblastoma. Enucleation was common treatment option. However, due to large size of the lesion, age, anatomical structures, and teeth involved, incisional biopsy was done followed by marsupialization after informed consent was obtained from the parent. Fine needle aspiration cytology was done using 5 ml syringe where a milky-yellow liquid content was obtained [Figure 2a]. Extraction of 84, 85 was performed and red wine-colored material appeared inside the cavity was collected [Figure 2b]. An impression was made followed by fabrication of thermoform surgical splint [Figure 2c], extending into surgical site with buccal and lingual flanges to support fragile walls from collapse [Figure 2d and e]. Parents were given oral hygiene instructions regarding maintenance of splint, and irrigation of socket with betadine, and saline thrice daily. The patient was explained regarding the treatment plan of 36, 74, but the patient was intended to get it done later as they were more worried about the existing lesion. Cytopathologic examination of the aspirate showed dense population of neutrophils, lymphocytes, foamy macrophages, few multinucleate giant cells and keratinocytes against hemorrhagic background suggestive of keratin filled lesion. Biopsy revealed parakeratinized stratified epithelial lining with prominent columnar basal cell layer, surface corrugation, and neutrophils, lymphocytes arranged against extravasated red blood cells [Figure 2f], which confirmed the diagnosis to be OKC. Follow-up was done every 3 days till 1st month which showed occlusal movement of developing tooth bud clinically, as well as reduction in radiolucency radiographically [Figure 2g]. Later, the patient was reviewed every 6 months till 3 years, in which there was a gradual reduction in radiolucency, occlusal movement of developing tooth buds and accordingly surgical splint was refabricated. During the 2nd-year follow-up, endodontic treatment was performed in 36 followed by composite inlay. By the end of 3 years, there was complete healing of the lesion with eruption of all permanent teeth [Figure 3a-f].

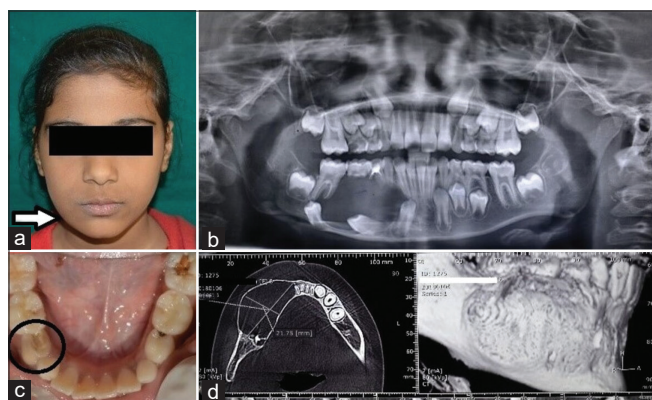


Figure 1: Preoperative pictures. (a) Extraoral swelling on right side of face (arrow) with facial asymmetry (b) Vestibular obliteration (circled) at 84, 85 (c) Orthopantomogram showing single homogenous radiolucency in the region of 83, 84, 85, 46 (d) Cone beam computed tomography showing expansion of buccal and lingual cortical plates

DISCUSSION

OKCs are common developmental odontogenic cysts occurring three times more often in the mandible than the maxilla and is most frequently seen in the region of canine to premolar, mandibular retromolar, ramus areas and maxillary second permanent molar area.^[3] It should be often differentiated from Dentigerous cyst, radicular cyst and lateral periodontal cyst which are other endodontic lesions.^[11] Radiographically OKCs display a radiolucency extending along the length of the mandible displacing developing teeth in children and adolescents. Histopathologically, OKCs are characterized by 5–8

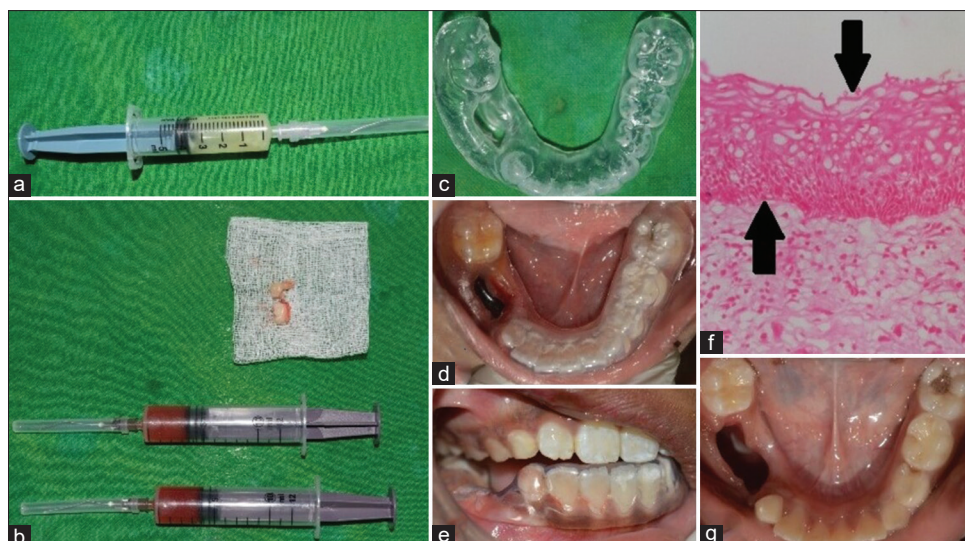


Figure 2: (a) Aspirated fluid (milky yellow in color) (b) Extracted 84, 85 and red wine-colored fluid collected (c) Fabricated thermoform surgical splint (d and e) Inserted marsupialization splint (f) Histologic picture showing parakeratinized stratified epithelial lining with prominent columnar basal cell layer (arrow), surface corrugation (arrow) (H&E stain, x40) (g) Follow-up after 1 month

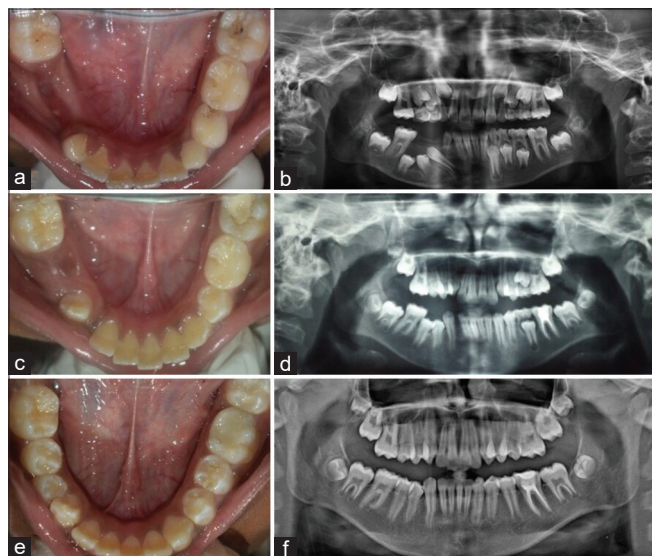


Figure 3: Postoperative clinical and radiographic follow-up showing remarkable reduction in lesion size and eruption of premolars (a and b) After 10 months (c and d) After 2 years (e and f) After 3 years

layered parakeratinized stratified squamous epithelium lining connective tissue capsule. Basal layer has palisade arrangement of hyperchromatic nuclei in columnar epithelial cells. The epithelial projections are absent in epithelial-connective tissue junction. The consistency of luminal content can be described as a “straw-colored fluid,” “thick pus-like” material or a caseous, cheesy, thick, milk-white mass which was similar in this case.^[5] Treatment of OKCs remains controversial because of their higher recurrence potential,^[12] and options include decompression, marginal resection, *en bloc* resection and adjuvant therapy, like cryotherapy, peripheral ostectomy and

Carnoy’s solution.^[2] However, marsupialization (Partsch I procedure)^[7] is reported to be the treatment of choice in large cysts involving multiple unerupted teeth and anatomic structures. It aims to reduce the size of the lesion thus preventing extensive surgery, preserving vital structures.^[9] In literature, the term marsupialization is often used interchangeably with decompression, but they are distinguished as decompression is any technique that reduces the pressure within a cyst, while marsupialization is a process of converting cyst into an open pouch that communicates with the oral cavity.^[7] However, both the procedures allow new bone to fill the defect. Studies have shown that more effective results were observed after marsupialization in lesions of the mandibular body than in ramus.^[13]

Despite having advantages, it has few limitations like prolonged healing period, discomfort to the patient, requirement of irrigation twice daily and weekly modification of surgical splint maintaining the cystic opening.^[7] Various devices such as tubes, gauzes, obturators and acrylic stent can be used for cystic window, but due to drawbacks like retention failure, increased porosity, irritating effect of residual monomer and difficulty in processing, usage of thermoformed appliances has gained high demand.^[14] In this case, thermoform splint was used which is a polyoxymethylene-based material made up of acetal resin. It is retained in an adequate position by negative pressure created intraorally. This obturator has advantages like easy removal to irrigate the cavity, nontoxic, nonallergic, flexible, resists wear and fracture, making it an ideal material of choice as surgical stent.^[14] Ziccardi *et al.*

used marsupialization stent along with space maintainer to preserve space for unerupted teeth.^[10] In the present case, thermoform marsupialization splint itself maintained space between existing dentition and allowed the displaced teeth to erupt in proper alignment without requirement of an additional space maintainer or orthodontic traction.

Literature has reported that recurrence rate of OKC varies from 35% to 50% and 21.4% among lesions treated by marsupialization.^[13] An immunohistochemistry study noted that after marsupialization, the epithelium thickness was increased, and the keratinized epithelium was changed to a hyperplastic and nonkeratinized stratified squamous epithelium.^[7] There is a histological transformation of 23 aggressive keratocysts to either less aggressive OKC or nonkeratocysts before and after marsupialization. Interleukin 1 and Ki-67 reduction was also observed in many cases, which is considered to be the factor for decrease in volume of the cysts^[15] allowing new bone to fill the defect.^[7] Thus, growth characteristics of OKC may become less aggressive during the course of marsupialization.^[13] implying that recurrence rate is not affected by it.^[7] According to Forssell *et al.*, an active follow-up performed annually during a 3-year period in pediatric patients might be sufficient, to ensure absence of recurrence and adequate eruption of the permanent teeth.^[13]

In conclusion, the diagnosis of OKC can be confirmed histologically along with radiological findings. In the selection of the most appropriate treatment, one must consider patient factors such as age, general medical condition and lesion characteristics. Considering the young age of the patient, subsequent growth of mandible and facial contour, treatment by marsupialization was performed in this case without any recurrence at 3-year follow-up which was an unique scenario. Although marsupialization is an unconventional method of treating OKC, it is a very effective method in children where there is proximity of lesion to the developing tooth buds and vital structures in the jaws. For the treatment success, regular follow-ups, along with patient awareness in maintaining oral hygiene, are important.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal.

The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, *et al.* Odontogenic keratocyst: Imaging features of a benign lesion with an aggressive behaviour. *Insights Imaging* 2018;9:883-97.
- Telles DC, Castro WH, Gomez RS, Souto GR, Mesquita RA. Morphometric evaluation of keratocystic odontogenic tumor before and after marsupialization. *Braz Oral Res* 2013;27:496-502.
- Ozkan L, Aksoy S, Orhan K, Cetiner S, Uyanik LO, Buhara O, *et al.* Case report: Multiple keratocystic odontogenic tumour in a non-syndromal pediatric patient. *Eur J Paediatr Dent* 2014;15:241-4.
- Madras J, Lapointe H. Keratocystic odontogenic tumour: Reclassification of the odontogenic keratocyst from cyst to tumour. *J Can Dent Assoc* 2008;74:165-165h.
- Rajkumar GC, Hemalatha M, Shashikala R, Sonal P. Massive keratocystic odontogenic tumor of mandible: A case report and review of literature. *Indian J Dent Res* 2011;22:181.
- Stoelting PJ. Long-term follow-up on keratocysts treated according to a defined protocol. *Int J Oral Maxillofac Surg* 2001;30:14-25.
- Briki S, Elleuch W, Karray F, Abdelmoula M, Tanoubi I. Cysts and tumors of the jaws treated by marsupialization: A description of 4 clinical cases. *J Clin Exp Dent* 2019;11:e565-9.
- Myoung H, Hong SP, Hong SD, Lee JI, Lim CY, Choung PH, *et al.* Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;91:328-33.
- Kim J, Nam E, Yoon S. Conservative management (marsupialization) of unicystic ameloblastoma: Literature review and a case report. *Maxillofac Plast Reconstr Surg* 2017;39:38.
- Wiemer SJ, Pruitt CA, Rallis DJ, Viozzi CF. Use of a modified removable partial denture as a marsupialization stent in a pediatric patient. *J Oral Maxillofac Surg* 2013;71:1382-6.
- Prashanth BR, Vidhya MS, Karale R, Kumar GV. Is odontogenic keratocyst an endodontic enigma? A rare case report of management of odontogenic keratocyst in anterior mandible. *J Oral Maxillofac Pathol* 2020;24:S7-10.
- Deboni MC, Brozoski MA, Traina AA, Acay RR, Naclério-Homem Mda G. Surgical management of dentigerous cyst and keratocystic odontogenic tumor in children: A conservative approach and 7-year follow-up. *J Appl Oral Sci* 2012;20:282-5.
- Nakamura N, Mitsuyasu T, Mitsuyasu Y, Taketomi T, Higuchi Y, Ohishi M. Marsupialization for odontogenic keratocysts: Long-term follow-up analysis of the effects and changes in growth characteristics. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;94:543-53.
- Checherita L, Beldiman MA, Stamatini O, Foia L, Forna NC. Aspects on structure of materials used for different types of occlusal splints. *Rev Chim* 2013;64:864-7.
- Tabrizi R, Özkan BT, Dehghani A, Langner NJ. Marsupialization as a treatment option for the odontogenic keratocyst. *J Craniofac Surg* 2012;23:e459-61.