



Conservative Treatment of Odontogenic Keratocyst Using a Specially Designed Acrylic Stent-Obturator: A Case Report

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ABSTRACT

An odontogenic keratocyst is a common oral lesion with high recurrence rates. There is an absence of a solid treatment consensus. Recurrence rates are reported to be higher in conservative treatment options. In this case report, we present a new method, utilizing a specially fabricated acrylic stent-obturator, aiding the conservative treatment methods in order to minimize lesion recurrence. A 42-year-old Saudi male with a 3.5 x 2.5 keratocyst on the right mandibular angle was treated successfully and conservatively in full abidance by medical community standards using the stent-obturator after lesion curettage alone, with no recurrence at three year follow up.

KEYWORDS

Odontogenic keratocyst, conservative management, obturator, re-curettage

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1. Introduction

The odontogenic keratocyst (OKC) has been described in the literature as an aggressive but benign odontogenic lesion contained within the bony structures of the jaw. Its origin is believed to be from the remnants of the dental lamina, as the name suggests, OKC forms a cystic space that could be occupied by keratin. Cystic lining is composed of multiple layers of parakeratinized squamous epithelium, with a basal layer that is formed from palisaded columnar cells or cuboidal cells type (Boffano *et al.*, 2021). The cyst has a tendency toward infiltrating the surrounding tissues by forming satellite cells in its basal layer (Borghesi *et al.*, 2018).

OKC can occur at almost any age, with some reports documenting OKC diagnoses in an eight-year-old child and up to an 82-year-old adult. However, the third decade of life registers OKC peak diagnosis with more incidence in the male population. OKC has also been reported in both the upper and lower jaw, with the maxilla bearing only half the number of mandibular cases; in particular, most cases were reported in the posterior mandible. A keratocyst could be manifested with dental structure; multiple OKC is usually one of the diagnosis criteria of Gorlin-Goltz syndrome (Borghesi *et al.*, 2018; de Castro *et al.*, 2018; Safadi *et al.*, 2020). OKC is also reported in no association with any dental structures (Al-Bodbaaj *et al.*, 2016). Moreover, some extensive tangling with deeper and more vital structures has been described in the literature (Goto *et al.*, 2020).

In regard to radiographic appearance, it can be seen as a unilocular or multilocular, well-defined, radiolucent lesion surrounded by a clear corticated margin that may cause resorption of surrounding teeth roots or expansion and perforation of the cortical plates (Borghesi *et al.*, 2018).

OKC management methods may include enucleation-curettage alone, marsupialization, or decompression with enucleation as conservative ways of treatment and up to marginal or en bloc resection as an aggressive way of treating the lesion (Boffano *et al.*, 2021).

2. Case Report

A 42-year-old Saudi male was referred to the oral and maxillofacial department at King Faisal Hospital, Al Ahsa, Saudi Arabia, for consultation and further investigation of a radiolucent lesion in the posterior region of the right side of the mandible, discovered during a routine dental examination.

The medical history of the patient was unremarkable. The extraoral and intraoral examination showed neither swelling nor color changes, and the patient reported no symptoms at all.

Orthopantomography showed a well-defined, radiolucent lesion in the posterior region of the right side of the mandible, with a scalloped and sclerotic border. The radiolucency extended from the distal surface of tooth number 47, which is separated from the lesion by a thin layer of bone, to approximately the middle of the ramus with no effect in the surrounding structure. The estimated dimensions of the lesion were 3.5 x 2.5 cm (see Figure 1). The patient has no wisdom teeth and does not recall any history of wisdom teeth extraction. Based on radiograph appearance, our differential diagnosis was odontogenic keratocyst, residual cyst and, less possible, unicystic ameloblastoma.

On 4 October 2018, fine needle aspiration and incisional biopsy was taken and sent to the pathology laboratory. The aspiration showed a straw-colored and cheesy fluid; laboratory analysis revealed few squamous cells in a background of granular keratinous debris, no protein estimate was carried out for the aspirate. The incisional biopsy revealed a cystic lesion lined by uniform, 6–10-thick, parakeratinized, stratified squamous epithelium, characterized by a corrugated surface, palisaded basal cell layer nuclei, and flat epithelium connective tissue interface (see Figure 2A). Based on this, our initial diagnosis of odontogenic keratocyst was confirmed.

After discussing the treatment options with the patient, the patient insisted on conservative treatment and refused to “walk with a tube or gauze in his mouth for months” (in patient’s own words). Thus, marsupialization or decompression options were excluded. We

discussed another conservative option with the patient involving using a specially designed acrylic stent-obturator to keep the lesion site open for re-curettage in order to reduce the recurrence rate.

Upon patient's approval on October 21, 2018, the lesion was deroofed, enucleated, and curettage performed on the dental chair under local anaesthesia (lidocaine hydrochloride 2% with epinephrine 1:80,000, inferior alveolar nerve block, one carpule). The removed tissue was sent for histopathologic examination. Additionally, an alginate impression was taken of the site of operation to construct the removable stent-obturator described above. The impression was sent to the lab and a diagnostic cast was made. A custom design stent-obturator was made on the diagnostic cast using a suck-down machine. The posterior part of the stent-obturator that was to occupy the cystic cavity was reinforced by orthodontic acrylic. Thirty minutes later, the stent-obturator was tried, adjusted in the clinic, and delivered to the patient (see Figures 3 and 4).

The histopathologic examination of the excisional tissue came in concordance with the incisional biopsy result, confirming OKC diagnosis.

Two weeks after the first visit, the lesion underwent re-curettage, and the removed tissue was sent to the histopathologic examination, which revealed only granulation tissue, without any evidence of OKC (see Figure 2B). In addition, the patient reported a slight numbness on the lower lip at the surgical sight. Two weeks' follow-up, on 6 November 2018, showed a clean operation site with good healing (see Figure 5). Two months' follow-up visit, on 19 December 2018, also showed a clean operated site with significant healing and normal lip sensation. At this visit, the patient was instructed to stop wearing the stent-obturator. Intraoral examination after four months, on 17 February 2019, showed complete healing of the operated site. The case has been followed for three years, until 24 October 2021, with no recurrence (see Figure 6).

Figure (1): Panoramic radiograph showing large radiolucency on the right mandibular angle



Figure (2): (A) H&E cross section showing a uniform, 6–10 thick, parakeratinized, stratified squamous epithelial characterized by a corrugated surface, palisaded basal cell layer nuclei, and flat epithelium connective tissue interface; (B) multiple fragments of granulation tissue with a piece of pre-existing bone.

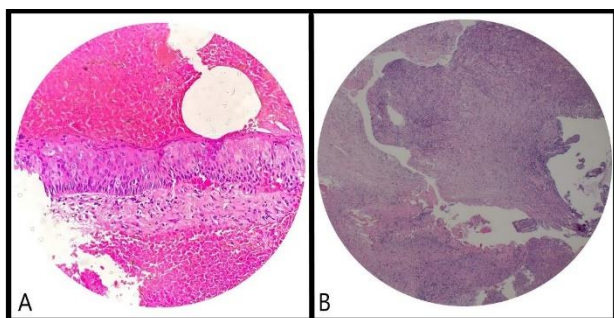


Figure (3): Showing the specially fabricated acrylic stent-obturator, reinforced part (arrow)



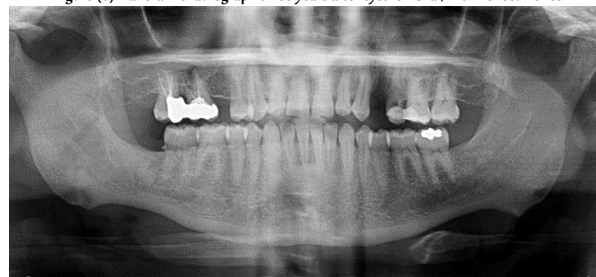
Figure (4): Showing the acrylic stent-obturator inside patient's mouth



Figure (5): Two weeks after cyst enucleation, and patient still wearing the stent-obturator



Figure (6): Panoramic radiograph three years after cyst removal, with no recurrence



3. Discussion

OKC is commonly approached via more conservative methods like marsupialization, decompression with or without enucleation, enucleation by itself, or enucleation with peripheral osteotomy, which is considered an aggressive adjunctive therapy. Although they show less recurrent rates, the more aggressive or radical methods like marginal resection are minimally used, making a conservative approach the preferable method for the surgeon (Boffano *et al.*, 2021; Mohanty *et al.*, 2021). Many factors affect the recurrence rate, including the size of the lesion, age of the patient, cortical perforation (which is associated with multilocular lesions), association with dentition and surrounding tissue infiltration (Titinchi, 2021).

Decompression accompanied by enucleation is a good treatment choice, especially if accompanied by peripheral osteotomy. This

combination is reported to significantly reduce the recurrence rate and is advocated in the OKC-suggested management protocols (Titinchi, 2020). On the other hand, utilizing conservative methods alone, such as simple enucleation, without adjunctive therapies like peripheral osteotomy and Carnoy's solution, shows a higher recurrence rate and therefore is not recommended (Zecha *et al.*, 2010). The use of chloroform in human beings has been restricted by the American Food and Drug Administration (FDA), which has led to avoidance of Carnoy's solution usage in this case (Helman *et al.*, 2007; Dashow *et al.*, 2013).

Post-operative infection is another issue to consider while treating jaw cysts; this was reported in a multi-center study of OKC (Boffano *et al.*, 2021). Accordingly, bacterial infection within the jaw cyst should be viewed cautiously, especially with new reports suggesting links between infection and lesion progression (Scalas *et al.*, 2013).

In this case, and in order to avoid infections and patient discomfort associated with marsupialization and decompression, and due to patient refusal of these methods, we used an acrylic stent-obturator, which served the purpose of keeping the operation site open yet clean in order to aid in re-curettage and minimize the recurrence rate. Due to repetitive curettage, and thanks to the stent-obturator, which served well in keeping the surgery site open for re-curettage and clean from debris or food accumulation, we also experienced increased patient comfort, avoidance of inflammation, and highly disturbing bad breath. Although the lesion has relatively small in size, we preferred to perform re-curettage over simple enucleation due to the higher recurrence rate associated with simple enucleation (Titinchi, 2020). To our knowledge, there are no reports documenting or reporting the use of acrylic stent-obtutors for the described method in treating OKC. We came across the use of an acrylic obturator in a dentigerous cyst's conservative decompression with reported successful outcome (Chouchene *et al.*, 2021). The presented case has been treated conservatively and successfully in full abidance to treatment protocols acknowledged by the medical community in such cases, with the new idea of using an acrylic splint and with no recurrence at the three-year follow up.

4. Conclusion

Based on the combination of patient comfort, expected reduced chances of infection, and setting the grounds for multiple curettages with minimal patient and surgical site disturbances aiming for recurrence reduction, we strongly advocate the repetition of our presented method in the conservative treatment of OKC or similar oral lesions when conservative treatment is advocated and decompression or marsupialization are, for one reason or another, not an option.

Biographies

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Dr. Al Ali is a Saudi senior resident in oral medicine and oral pathology in the Saudi board program, with a bachelor's in dentistry from the University of Dammam, Saudi Arabia, 2010. Has training at Prince Sultan Military Hospital, Riyadh, Saudi Arabia. Research interests include management of medically compromised patients and effects of drugs on dental treatment. Has attended multiple dentistry-related conferences and courses and has multiple publications and scientific posters.

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