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Long-term observation of a large keratocystic odontogenic tumour of the mandible treated by a single enucleation procedure: A case report and literature review



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ABSTRACT

INTRODUCTION: Keratocystic odontogenic tumours (KCOTs) are benign lesions of the jaw that are characterised by expansive growth and high rates of recurrence. Herein, we present a novel minimally invasive method for the surgical treatment of KCOTs.

CASE PRESENTATION: We present a 49-year-old woman with a rare, large KCOT of the mandible extending from tooth 37 to tooth 47. A single enucleation procedure was selected as the surgical technique combined with a titanium plate. Teeth interfering with the cystic lumen were preserved. The bone surrounding the cyst was partially removed and rinsed with Carnoy's solution. Recurrence was observed 1 year later between teeth 43 and 45, and was treated by single enucleation with Carnoy's solution. After the second operation, the teeth interfering with the KCOT still elicited a positive response to dental pulp testing. No further signs of recurrence were observed after a total observation period of 7 years.

DISCUSSION: Various surgical interventions have been described for KCOTs. However, a general guideline for a specific surgical intervention has not yet been established owing to the heterogeneity of these tumours.

CONCLUSION: Immediate enucleation in combination with a titanium plate offers an effective surgical approach for the treatment of large KCOTs of the mandible without the need for radical tooth removal or resection of the afflicted side.

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1. Introduction

Keratocystic odontogenic tumours (KCOTs) were first described in 1956 as benign neoplasms of the jaw that have a high risk of recurrence [1]. Considering their aggressive infiltrative behaviour that often leads to penetration of the surrounding tissues, KCOTs exhibit the combined characteristics of cysts and benign tumours [1,2]. KCOTs are usually characterised by an 8–10 cell thick parakeratinised epithelium and represent 12–14% of all odontogenic cysts of the jaw [1,3]. Clinical symptoms of odontogenic keratocysts are rarely observed; they are often discovered incidentally through radiographic examination [4]. KCOTs generally present as

well-defined radiolucent lesions with a radiopaque cortical margin [5].

In most instances, when the KCOT grows to a large size, marsupialisation or decompression with subsequent enucleation is performed to reduce the size of the tumour and preserve adjacent structures [6].

Herein, we present a case involving a large KCOT of the mandible treated by enucleation with Carnoy's solution (CS), peripheral ostectomy, and a 2.0-mm 16-hole titanium plate. Teeth interfering with the cystic lesion were not extracted, but still elicited a positive response to dental pulp testing after the operation. This case report is compliant with the SCARE guidelines [7].

2. Case presentation

In May 2009, a 42-year-old woman developed a sudden, painless swelling in the posterior region of the right side of her mandible that continued for approximately 14 days. The patient had experienced neither pathological mobility of the teeth nor malocclusion. Her medical and family histories were unremarkable. A panoramic radiographic (PR) examination had not been previously performed. Subsequent PR imaging revealed a large, radiolucent, expanding

Abbreviations: CT, computed tomography; CS, Carnoy's solution; KCOT, keratocystic odontogenic tumour; PR, panoramic radiograph.

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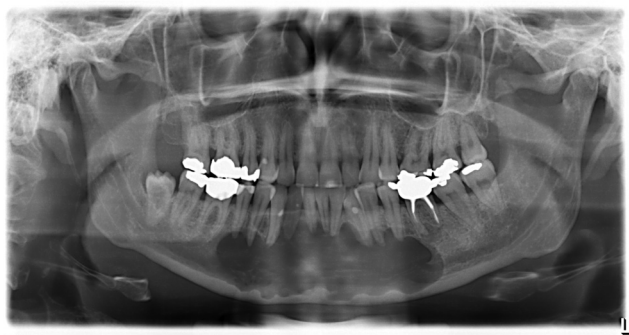


Fig. 1. Pre-surgical panoramic radiographic image (2009).

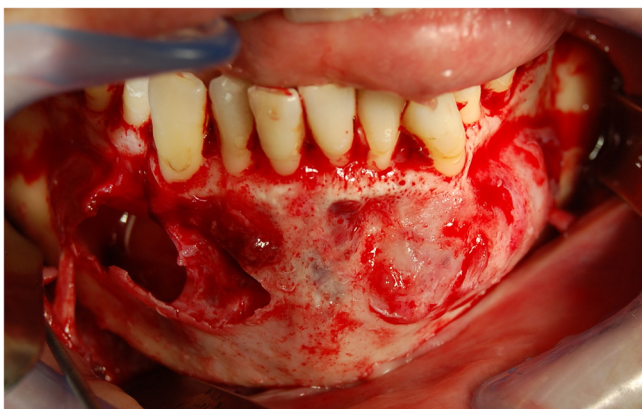


Fig. 2. Intra-oral view of the cystic lesion during surgery with perforation of the cortical bone (2009).

lesion involving the majority of the mandibular bone from the distal side of tooth 35 to the distal side of tooth 45 with no evidence of root resorption (Fig. 1). An axial computed tomography (CT) scan revealed a well-defined multilocular cystic lesion. On the right side, a perforation in the cortical region was examined (approximately 10 mm in diameter) with the overlying soft tissue. Biopsy findings were suggestive of a KCOT.

Surgery was performed under nasotracheal intubation. A mucoperiosteal flap extending from teeth 37–47 was prepared. Exceedingly thin layers of compact bone covering the cystic lumen were removed (Fig. 2). The cyst was enucleated by removing the surrounding bone. Owing to the large size of the KCOT, and to reduce the risk of a mandibular fracture, a 2.0-mm titanium plate (MatrixMANDIBLE Plating System®; Synthes CMF, West Chester, PA, USA), fixed with 3- × 10-mm screws on each side, was installed (Fig. 3). The cystic defect and the area surrounding the roots of the affected teeth were carefully swabbed with CS (60% ethanol, 30% chloroform, and 10% glacial acetic acid) as a curative agent for only 1 min to avoid damaging the nerves around the teeth, followed by irrigation with sodium chloride. The defect was then closed using a 5-0 PROLENE® polypropylene suture. Histological analysis of the specimen confirmed a final diagnosis of a parakeratinised KCOT.

After the initial surgery, the patient underwent clinical examinations every 3 months and PR and CT examinations every 6 months. In August 2010, a recurrence of the KCOT was suspected based on PR and CT findings. A CT scan revealed three radiolucent regions in the mandibular bone extending from the mesial side of tooth 43 to the distal side of tooth 45 (Fig. 4). A second operation was performed under nasotracheal intubation. Three cystic lesions were enucleated, the surrounding bone was partially removed, and CS was applied for 1 min followed by irrigation with sodium

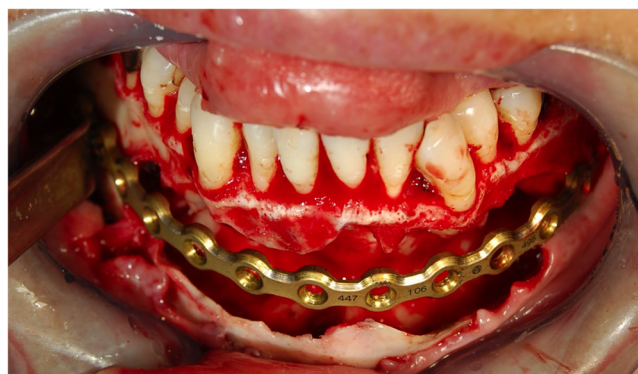


Fig. 3. Intra-operative placement of the 2.0 mm 16-hole titanium plate (MatrixMANDIBLE Plating System®; Synthes CMF, West Chester, PA, USA) (2009).



Fig. 4. Pre-surgical computed tomography image showing three recurrent cystic lesions (2010).

chloride, as in the first operation. Histological analysis of the specimen confirmed a diagnosis of a recurrent KCOT. No complications occurred during surgery, and post-surgical healing was satisfactory.

Clinical follow-up examinations with PR and CT scans were performed for early detection of any recurrence every 6 months until March 2016; no signs of recurrence were observed (Fig. 5).

3. Discussion

KCOTs are benign lesions of the jaw that frequently expand into the medullary cavity in the anteroposterior direction without perforating the solid bone [8,9]. Rapid growth of KCOTs may occur because of an active epithelial lining with a high rate of proliferation [5]. In our patient, the KCOT was located in the posterior region of the mandible on each side, as well as in the symphysis and premolar regions. Such KCOTs were reported in only 1.8% of 107 cases in a previous study [10]. Our patient noticed only a slight swelling

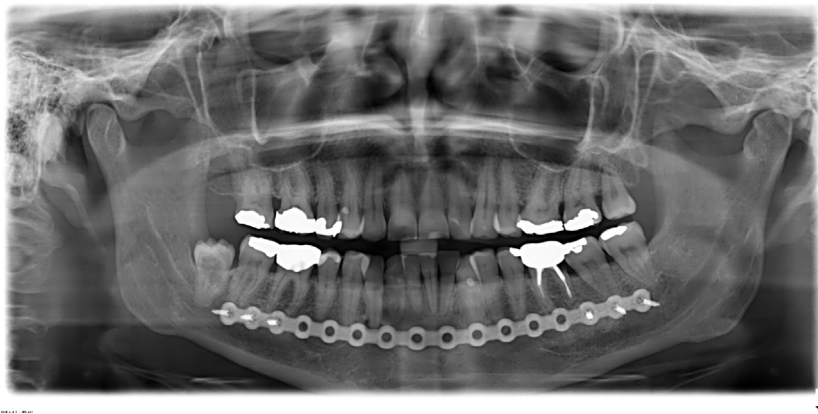


Fig. 5. Panoramic radiographic image demonstrating bone formation after a 7-year observation period (2016).

on the right side of the mandible, which probably appeared owing to an interrupted cortical bone structure.

Various treatment options have been described for KCOTs. The recurrence rates following surgical resection (1.9%) or enucleation with CS, or following decompression with subsequent enucleation (1.0–8.7%), were significantly lower than those following solid enucleation (17.0–56.0%) [6,11]. Dashow et al. reported a 7-fold increase in the recurrence rate of KCOTs treated with modified CS without chloroform compared to KCOTs treated with standard CS, as in the present case [12]. Another point of concern is the potential occurrence of temporary damages to the alveolar inferior nerve if CS is applied directly to nerve structures [13]. In a previous study, 25 of 83 patients experienced an inferior alveolar nerve deficit; 21 of these 25 patients experienced recovery of sensitivity after a mean time of 4.6 months [14].

Following each of the surgeries, the patient experienced transient paraesthesia of the right lower lip that gradually resolved within a few weeks. Teeth in the frontal region, extending from teeth 35–45, elicited a positive response to dental pulp testing that involved applying dry ice (solid carbon dioxide) to each tooth to elicit a response. As no other pathologies were detected, there was no need for further endodontic treatment. Recurrences following radical surgical procedures of various extents have been described in the literature¹⁵; therefore, it remains unclear whether tooth removal is in fact necessary. In our case, a satisfactory intraoperative assessment of the surgical site and achievement of macroscopic simple enucleation of the afflicted bone, combined with the young age of the patient and positive vitality of the teeth, helped us decide to pursue the path of minimally disruptive enucleation without removing the teeth. Different treatment options may also be considered to support teeth preservation [1]. For example, marsupialisation (which is primarily used for large KCOTs) can reduce damage to anatomical structures and may facilitate new bone formation to fill the defect by relieving intracystic pressure, enabling the cavity to gradually decrease in size [1]. However, only instruction-compliant patients are eligible for this procedure as it requires irrigation of the exposed lesion over a sufficient number of months to prevent infection [16].

In the present case, we used a titanium plate to reduce the risk of mandibular fracture. Although teeth were observed within the cyst cavity, they were preserved. KCOTs extending to nearby structures (e.g., overlying tissue or affected teeth) must be removed owing to the difficulty of enucleating potential adherent rests from the cyst [15] and to reduce the likelihood of recurrence [2,17–19]. In the present case, recurrence was likely caused by residual epithelial islands in the tissue overlying the cystic lesion, which was not removed. This was probably the result of an interrupted bone structure on the right side. In another study, the teeth interfering with

the cystic lesion were either extracted or treated by apicoectomy; however, the patient still experienced recurrence [15]. Because of the large expansion of the KCOT in our case, a radical surgical intervention was justified. An even more radical intervention could have reduced the likelihood of recurrence [20]; we observed recurrence after 1 year of follow-up. However, even recurrences in bone grafts were observed, which may not justify the morbidity associated with such a radical surgical intervention [15,17]. Therefore, immediate enucleation with CS and placement of a titanium plate may represent a convenient and effective treatment for large KCOTs. More radical interventions could be implemented in cases of multiple recurrences, despite life-long follow-up being mandatory.

4. Conclusions

We presented an effective and minimally invasive method for the treatment of KCOTs that successfully preserves anatomical structures and maintains the patient's quality of life. Compared to other methods (e.g. marsupialisation and decompression) with subsequent enucleation, a second operation can generally be forgone, as can months-long irrigation of the cystic lumen. Consequently, immediate enucleation in combination with a titanium plate offers an effective surgical option for the treatment of large KCOTs of the mandible without the need for radical tooth removal or resection of the afflicted side.

Conflicts of interest

None.

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Ethical approval

Not applicable. The present study is not a research study.

Consent

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Authors' contribution

Tobias L. Pittl provided clinical care to the patient, wrote the manuscript, and coordinated all aspects of this report. Dritan Turhani performed the surgery on the patient and provided clinical, radiological, and photographic documentation. Meier Marius, Walter Sutter, and Paul Hakl performed clinical and radiological follow-up treatment. All authors read and approved the final manuscript.

Guarantors

Dritan Turhani, Tobias L. Pittl.

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