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An odontogenic keratocyst treated 60 years later, via a sagittal split approach

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Abstract

Odontogenic keratocysts (OKC) are unique epithelium-lined developmental cysts of the jaws due to their greater growth potential, high recurrence rates and possible association with nevoid basal cell carcinoma syndrome. The gold standard for the treatment of OKCs is still debated, but it is known that adequate surgical access and success of the first enucleation is a predictor of non-recurrence. Sagittal split osteotomy (SSO) is an accepted surgical approach for lesions in the posterior mandible with the advantages of reducing cortical bone loss, a well-secured surgical field and reduced risk of injuring the inferior alveolar nerve or fracturing the mandible. A follow-up period of 10 years is currently accepted for OKCs.

This case report describes a 76-year-old male patient with a 60-year recurrence of an OKC in the right posterior mandible. The latest enucleation is the seventh attempt after previous procedures in the United States of America. The sixth attempt resulted in mental nerve severance and pathological fracture.

The report describes the successful removal of this lesion via a unilateral SSO approach and challenges the 10-year follow-up period for OKCs.

Introduction

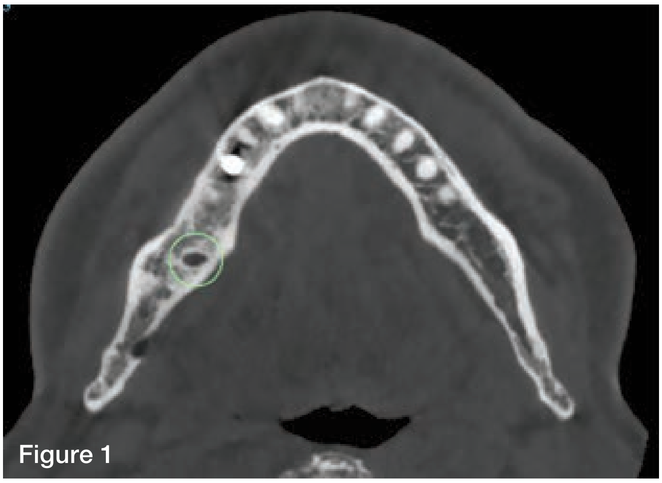
Odontogenic keratocysts (OKC), are epithelium-lined developmental cysts of the jaws originating from cell rests of the dental lamina (Neville et al., 2016; Phillipsen, 1956). They were previously known as “keratocystic odontogenic tumors” (KCOTs) prior to their reclassification from “tumours” by the World Health Organisation in 2017, however the names are still used synonymously in the literature. OKCs are unique to other developmental cyst of the jaws due to a greater growth potential, high recurrence rates and possible association with nevoid basal cell carcinoma syndrome (Neville et al., 2016). These cysts may be locally destructive, causing tooth root resorption (Chrcanovic and Gomez, 2017). Unlike dentigerous or radicular cysts, growth regulation is thought to occur through genetic factors or enzymic activity, rather than increasing osmotic pressure (Neville et al., 2016). OKCs contribute to 3-11% of odontogenic cysts (Chuong et al., 1982; Brannon, 1976; Ahlfors et al., 1984; Payne, 1972) and are found in patients of all ages, though most are diagnosed between the ages of 10-40 years with a slight male predilection. The majority of lesions are located in the posterior body and ramus of the mandible (60-80%) and may be unilocular or multilocular. These are often asymptomatic, however large lesions and particularly those breaching the bone cortices, may cause pain, swelling and drainage of contents into the oral cavity (Neville et al., 2016).

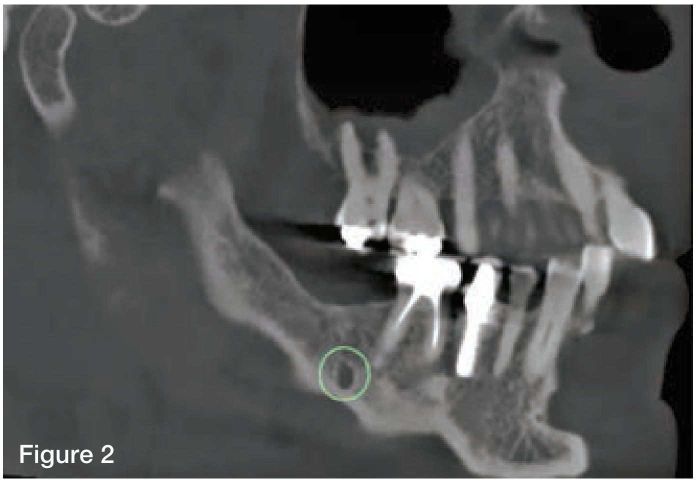
The gold standard for treatment and follow-up of OKCs is still debated. Accepted surgical modalities include marsupialisation/decompression, enucleation with or without adjunctive therapy (cryotherapy, Carnoy’s, peripheral ostectomy) and marginal/*en bloc* resection (Chrcanovic and Gomez, 2017). Intra-oral access to lesions of the posterior mandible may be via the buccal cortex, lingual cortex or using a unilateral sagittal split osteotomy (SSO). Extraoral approaches include submandibular or retromandibular incisions (Casap et al., 2006). Recurrence rates after surgical removal have been reported up to 62.5% (Pindborg and Hansen, 1963; Rud and Pindborg, 1969; Toller 1967), the average being 29.3% (Jung et al., 2021). The same study supports a 10-year duration of active annual follow-up after enucleation. A meta-analysis involving 6427 cases across 94 retrospective studies found that follow-up periods varied greatly between surgical centres with the latest recurrence recorded at 41 years (Crowley et al., 1992).

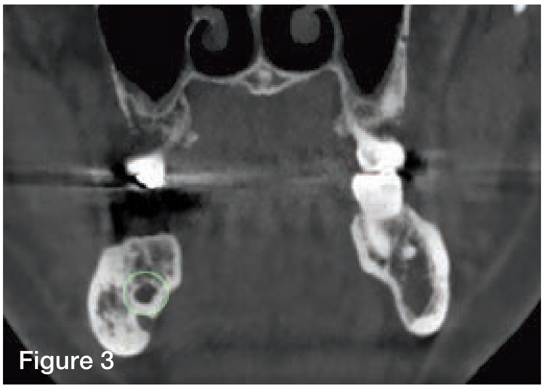
The following report demonstrates a recent case of OKC recurrence 60 years after initial surgical management as well as describing the successful removal of this lesion via an SSO approach.

Case report

A 76-year-old male was referred to the Oral and Maxillofacial Surgery (OMFS) unit at Christchurch Hospital, Canterbury District Health Board, New Zealand, in regard to a lesion in the right posterior body of the mandible, suspected to be a recurrence of a previously enucleated OKC. The history of the treated lesion was a personal account from the patient who was unable to remember which surgical centres in the USA he had visited. The historic documentation was therefore unable to be obtained and had been destroyed due to the length of time. The patient’s father, a radiographer, had reportedly shown him his radiograph with a large radiolucency in the posterior mandible extending up the ramus (the patient pointed to the right body and ramus of the mandible on current imaging when describing this). The lesion was first removed when the patient was 17 years of age, followed by a further five attempts. After the sixth attempt, 13 years prior, he was informed he had sustained right mental nerve severance and required nerve repair intra-operatively. Prior to our intervention, the patient reported the return of “most”







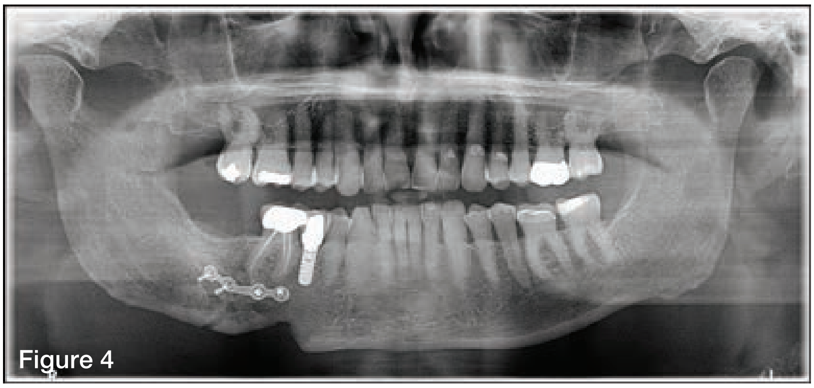


Figure 1. CBCT axial view showing a radiolucent lesion situated adjacent to the right lingual cortex of the mandible.
Figure 2. CBCT sagittal view showing a radiolucent lesion in the right hemi-mandible. Remodelling of the mandible can be seen by the irregularity of the inferior border below the second premolar implant.
Figure 3. CBCT coronal view showing a radiolucent lesion situated adjacent to the lingual mandibular cortex and superior to the inferior alveolar nerve canal.
Figure 4. Day 1, post-operative OPG showing anatomical reduction of the SSO and placement of a titanium mini-plate and screws.

of the sensation to the right lower lip and chin. Of note, a pathological fracture in the right body of the mandible was also sustained soon after the sixth enucleation attempt. This was discovered late and managed conservatively.

A cone beam computer tomography (CBCT) scan, taken by the current referrer during implant planning, incidentally demonstrated a 5 mm cystic lesion within the right mandible at the location of the extracted second molar (Figures 1, 2 and 3). This was monitored for 12 months and reported as having increased in size by a radiologist.

The lesion was surgically accessed through a unilateral SSO approach under general anaesthetic and found within the split. It was subsequently enucleated followed by peripheral ostectomy. Oragraft 70/30® (70% mineralised ground cortical bone with 30% demineralised ground cortical bone), allograft material, was packed into the cavity before reduction and fixation. Eyelets and wires were used to maintain the occlusion during the placement of a single 1.0 mm 4-hole Synthes® titanium mini-plate and 6 mm screws. An orthopantogram (OPG) was taken 24 hours post-operatively showing anatomical reduction of the osteotomy segments (Figure 4). The patient was reviewed three weeks later, reporting an uneventful recovery. He reported no gross changes to the right mental nerve sensation post-operatively. Ongoing annual clinical review with OPG and/or CBCT imaging will be pursued for at least 10 years. This will be followed by recommending 24-monthly imaging by the patient's general dentist.

The specimen was sent for histopathology in a 10% formalin solution and was reported by the hospital Anatomical Pathology team as an "odontogenic keratocyst" (Figures 5-8). The report noted "Calcified fragments and fibrovascular connective tissue lined by stratified squamous epithelium. The epithelium has a uniform basal layer and luminal surface of parakeratin showing focal corrugation. Focal budding growth from the basal cells and areas of subepithelial clefting are seen. The stroma contains occasional epithelial rests and patchy chronic inflammation".

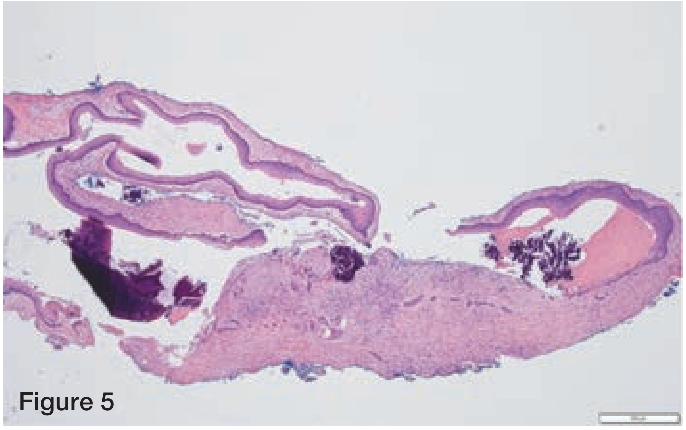
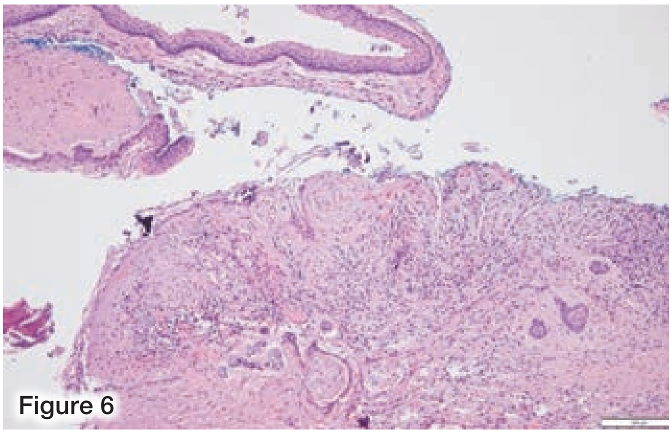


Figure 5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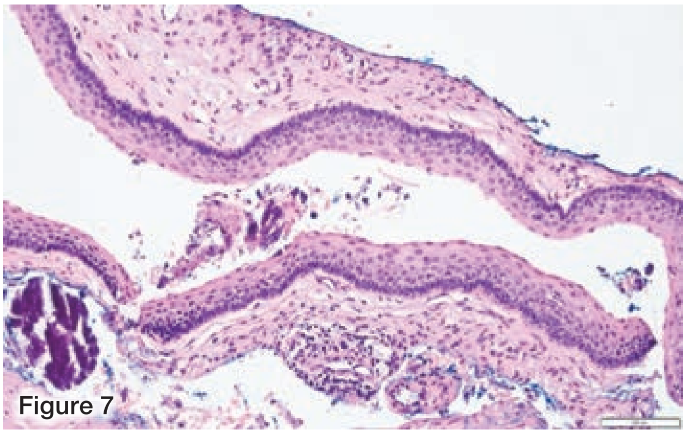
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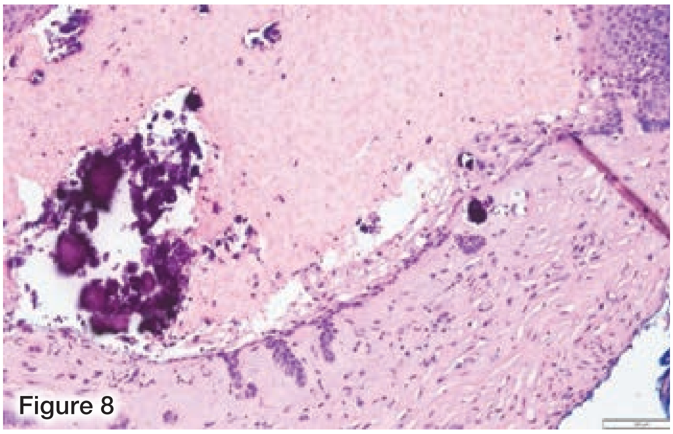
Figure 8

Low power magnification of the lesion with haematoxylin and eosin staining at 4X (Figure 5), 10X (Figure 6), 20X (Figure 7) and 40X (Figure 8).

Discussion

Recurrence of an OKC is more likely when there is inadequate surgical access during the initial operation (Bramley, 1974). The documented use of the SSO approach for mandibular pathology describes advantages such as ease of securing the surgical field, sparing of cortical bone, hastening recovery, reduced risk of injuring the inferior alveolar nerve, and minimisation of mandibular fractures (Lee et al., 2015; Casap et al., 2006). The authors found the use of this technique beneficial for direct access and visualisation of the relatively small lesion, simultaneously producing ideal access for the peripheral ostectomy and bone grafting. A sagittal split allowed for identification of the 5 mm cystic lesion along its length, whereas a standard buccal approach may have resulted in broad, deep exploration with unnecessary sacrifice of unaffected bone. A limitation of the SSO approach is its technique sensitivity and it may be considered a more "radical" approach compared to buccal cortical access. The treating team regularly perform osteotomies for orthognathic treatment and thus were comfortable proceeding with this approach for removal of the lesion. However, those who do not perform osteotomies as part of their usual practice may consider referral or an alternative approach for similar cases. Other limitations include the need for general anaesthetic and risk of complications such as infected metal-ware, non-union, alveolar/lingual nerve injury and unfavourable splits. Navigation technology, such as Stealth® or 3-dimensional printed bio-models with surgical guides are other options to be considered.

A recurrence later in life increases the chance of age-related complications, such as advanced general comorbidities, reduced oral rehabilitation options or reduced patient motivation for definitive treatment. Successful management at a young age both improves surgical safety and reduces recovery burden. In the current case, follow-up between this current and last enucleation was 13 years. While the chance of identifying a recurrence is highest in the first 10 years, there may be a place for prolonged radiographic follow-up beyond this timeframe. One might consider, for example, ongoing 24-monthly imaging beyond the initial 10-year annual reviews, on a case-by-case basis. The duration could be based on the size and behaviour of the original lesion knowing that recurrence is more likely in large or multiloculated lesions, or those invaginated between tooth roots. Extending the currently accepted follow-up period cannot be determined by a single case report, however this case will add to current literature and may encourage future robust clinical studies. Notably, a prolonged review period does mean increased patient cost, travel and exposure to radiation. However, the long-term benefit of early identification and enucleation of a small recurrence are likely to outweigh the risks of managing a recurrence at an older age or when the lesion is larger. Radiographic follow-up beyond the

10-year period may coincide with the patient's routine dental assessments, and with purposeful communication between the surgeon and the patient's regular dentist, this could minimise patient burden.

Conclusion

Adequate surgical access and prolonged follow-up is necessary for appropriate management of OKCs. The SSO approach can be used for patients with pathology in the posterior mandible while avoiding unnecessary bone removal and providing excellent direct vision. As demonstrated by the case presented, OKCs can recur 60 years after initial enucleation and hereby challenges the currently accepted cessation of active follow-up at 10 years. As a surgical specialist, the emphasis should always be to provide patients with the best care possible. By choosing the most appropriate surgical approach and recalling patients accordingly, the burden of managing recurrences may be minimised.

References

Ahlfors E, Larsson A, Sjogren S (1984). The odontogenic keratocyst: a benign cystic tumor? *J Oral Maxillofac Surg* 42: 10-19.

Bramley P (1974). The odontogenic keratocyst, an approach to treatment. *Int J Oral Surg* 3: 337.

Casap N, Zeltser R, Abu-Tair J, Shteyer A (2006). Removal of a large odontoma by sagittal split osteotomy. *J Oral Maxillofac Surg* 64: 1833-1836.

Chrcanovic BR, Gomez RS (2017). Recurrence probability for keratocystic odontogenic tumors: An analysis of 6427 cases. *J Cranio Maxillofac Surg* 45: 244-251.

Chuong R, Donoff RB, Guralnick W (1982). The odontogenic kerato- cyst. *J Oral Maxillofac Surg* 40: 797-802.

Crowley TE, Kaugars GE, Gunsolley JC (1992). Odontogenic keratocysts: a clinical and histologic comparison of the parakeratin and orthokeratin variants. *J Oral Maxillofac Surg* 50: 22-26.

Jung, HD., Lim, JH., Kim, H.J (2021). Appropriate follow-up period for odontogenic keratocyst: a retrospective study. *Maxillofac Plast Reconstr Surg* 43: 16.

Neville BW, Damm DD, Allen CM, Chi AC (2016). Odontogenic cysts and tumors. *Oral and Maxillofacial Pathology. Fourth edition.* Missouri: Elsevier (pages 632-689).

Pindborg JJ, Hansen J (1963). Studies on odontogenic cyst epithelium. 2. Clinical and roentgenologic aspects of odontogenic keratocysts. *Acta Pathol Microbiol Scand* 58: 283-294.

Rittersman J, van Gool AV (1979). Surgical access to multicystic lesions, by sagittal splitting of the lower jaw. *J Maxillofac Surg* 7: 246.

Rud J, Pindborg JJ (1969). Odontogenic keratocysts: a follow-up study of 21 cases. *J Oral Surg* 27: 323-330.

Toller P (1967). Origin and growth of cysts of the jaws. *Ann R Coll Surg Engl* 40: 306-336.

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