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Odontogenic Keratocyst: A Case Report Emphasizing on Root Canal Treatment after Surgical Intervention

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ABSTRACT

This article presents a case of odontogenic keratocyst (OKC) located in the mandible, involving teeth 36 to 45, with significant loss of alveolar bone and aseptic pulp necrosis, emphasizing on root canal treatment after surgical intervention. Orthopantomogram and computed tomography examinations revealed an extensive, well-defined, and multilocular radiolucent lesion. Histopathological examination after incisional biopsy confirmed OKC, which was removed completely with enucleation and curettage, followed by the endodontic treatments of teeth 36 to 45 using reciprocating nickel-titanium files (Reciproc) in a single session. Afterwards, teeth 33 to 36 underwent apical surgery to create an appropriate bone development. Panoramic radiographic images showed bone formation and no sign of recurrence after one-year follow-up. In conclusion, this surgical approach, combined with the endodontic treatments of the teeth involved in the lesion, was effective for the management of OKC, promoting injury regression and preservation of the natural teeth.

Keywords: Dental Pulp Necrosis; Enucleation; Odontogenic Cysts; Odontogenic Keratocyst; Root Canal Therapy

Introduction

Odontogenic Keratocyst (OKC), previously called keratocystic odontogenic tumour, is a benign intraosseous odontogenic lesion, presented as either an isolated form or multiple lesions in the same individual. The latter is usually associated with the nevoid basal cell carcinoma syndrome (NBCCS) or Gorlin-Goltz syndrome, which is an autosomal dominant multi-systemic disease [1]. Its development arises from epithelial remnants of dental lamina (epithelial lining of the developing tooth follicle) or offshoots of the basal cell layer of the oral epithelium, nevertheless, the triggering factors are still unclear [2]. Microscopically, OKC has a characteristic lining of parakeratinised stratified squamous epithelium exhibiting a palisaded basal layer with basophilic nuclei and a potentially aggressive and infiltrative behavior [1, 3, 4].

Odontogenic Keratocyst represent approximately 10% of all jaw cysts [5]. OKCs also show a high recurrence rate (16.6% to 21.1%) particularly in patients with multiple lesions which indicates their potential for malignancy [1, 6, 7]. OKCs may cause intense and rapid bone destruction and reach large dimensions, often without particular symptoms and significant bone deformation [6]. Radiographically, OKCs are presented as unilocular or multilocular radiolucent images with well-defined borders [1, 2]. The treatment of OKCs is complex and generally requires a multidisciplinary approach and multiple interventions that include marsupialisation (also called decompression), enucleation with or without adjunctive therapy (curettage, chemical cauterisation, cryotherapy, electrocautery and peripheral osteoectomy), and resection [1, 2, 7].

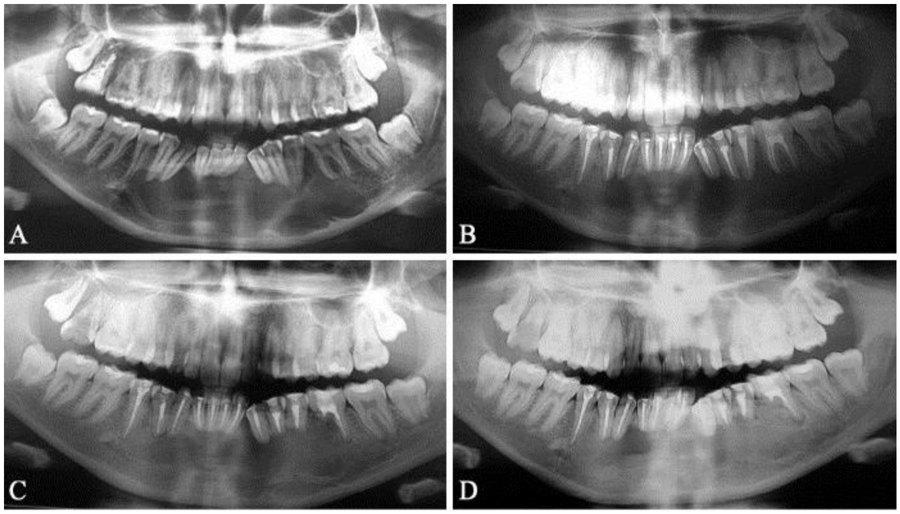

Figure 1. Orthopantomogram of the mandible: *A)* Pre-operative view (May 2017); *B)* One month after endodontic treatments (September 2017); *C)* One month after apicoectomy of teeth 33 to 36 (October 2017); *D)* One-year follow-up (September 2018)

Because of their periapical location, particularly when presented as a unilocular lesion, OKCs may sometimes be confused with pulp necrosis sequelae. Thus, in such cases, endodontic treatment alone may be unnecessary and will have no effect on the regression of the injury [3, 8]. Kontogiannis *et al.* [8] conducted a retrospective study with 1521 biopsies of periapical lesions with clinical diagnosis suggestive of pulp necrosis sequelae and found an average of 3.42% of cases diagnosed histologically as non-endodontic lesions, with a higher frequency of OKC (34.62%). In some cases, OKCs may cause extreme tissue destruction and affect the vascular system and nutrition support of the pulp because of the lesion's expansion or the need for a more radical treatment [9]. This draws the attention of endodontists to the possibility of OKCs occurring in the periapical region.

To the best of our knowledge, previous studies have not addressed the postoperative endodontic management of teeth involved in OKCs. This article presents a case of an OKC located in the mandible, involving teeth 36 to 45, with significant loss of alveolar bone and aseptic pulp necrosis, emphasizing on root canal treatment after surgical intervention. The diagnosis procedure, treatment protocol, and clinicopathological aspects are also discussed. This case report followed the SCARE 2018 guidelines [10].

Case Report

An 18-year-old female patient was referred to the Dental Specialty Centre of the city of Bragança Paulista, SP, Brazil, in May 2017. She complained about a swelling in the bottom region of the gingivolabial groove of teeth 33 to 36, which had increased significantly over the last six months. Based on a detailed review of the patient's medical and dental history, no systemic diseases were reported and the oral condition was contributory. The patient did not report any presence of spontaneous pain and clinical examination of teeth 36 to 45 showed negative response to percussion, palpation, and pulp sensitivity tests. The absence of pathological mobility of the teeth and the presence of crowding in mandibular anterior teeth were also observed. There was no evidence of pulp chamber exposure to the oral cavity, caries communicating to the pulp chamber, previous endodontic treatments, and presence of cracks and calcifications; which were confirmed radiographically. Thus, the pulp status was diagnosed as aseptic pulp necrosis.

An orthopantomogram revealed an extensive, well-defined, and multilocular radiolucent lesion in the mandible involving teeth 36 to 45 (Figure 1A); suggestive of ameloblastoma, odontogenic Myxoma, or odontogenic keratocyst. A cone-beam computed tomography (CBCT) was performed, which clearly showed the extent of the lesion from the disto-apical portion of tooth 36 to the disto-apical portion of tooth 45, and from the base to the alveolar process of the mandible (Figures 2A to D). The CBCT showed expansion of the buccal cortical bone with areas of loss of continuity, expansion of the lingual cortical bone between teeth 32 and 33, external root resorption in teeth 31 to 36, 42, and 44, and root divergence of teeth 32 and 33 (Figures 2A to D).

After obtaining informed consent about the treatment procedure of the patient, an incisional biopsy was performed under local anaesthesia for the differential diagnosis by

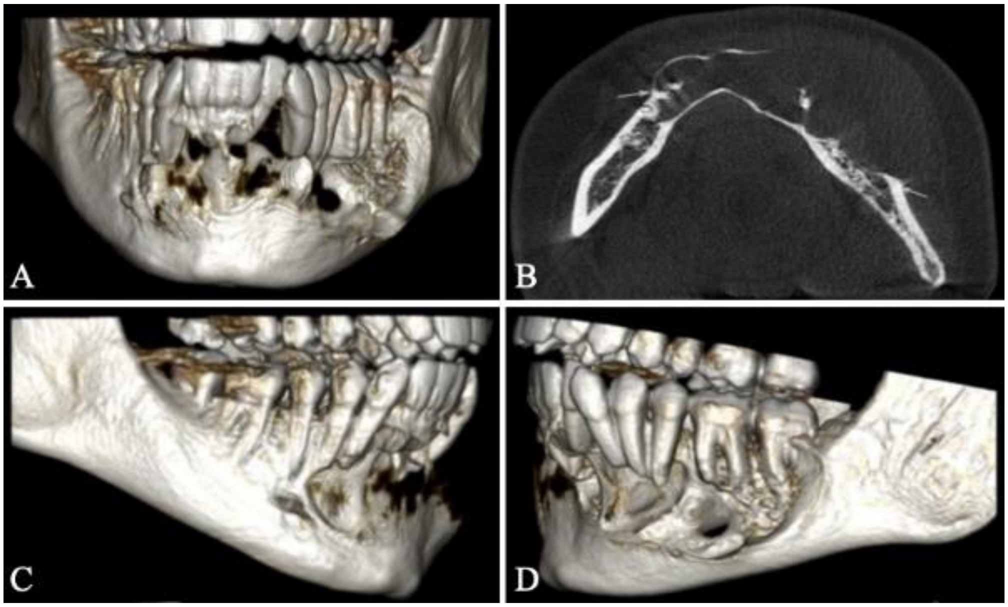

Figure 2. Pre-operative cone-beam computed tomography (CBCT) images of the mandible (May 2017): *A)* 3D front view; *B)* Axial view; *C)* 3D right side view; *D)* 3D left side view

histopathological examination. This examination confirmed the suspicion of odontogenic keratocyst (OKC), whose histological features were stratified epithelial basal cells with hyperchromatic parakeratinisation (Figure 3A). Considering the type and size of the lesion, different treatment protocols were discussed with the patient, who agreed to the complete exeresis of the lesion by enucleation and bone curettage, thus avoiding potential remnants of the lesion in the affected area (Figures 3B and C). Surgical intervention was completed four weeks after the initial visit. Due to the extent of the lesion and the amount of removed tissue, the patient was informed about the need for apicoectomy of teeth 33 to 36 after endodontic treatment. The healing of the surgical wound was uneventful and, one month after surgery, root canal treatments of teeth 36 to 45 were completed in five visits, with an average of two teeth treated at each visit.

A single operator treated all teeth endodontically in a single session, under local anaesthesia and absolute isolation with a rubber dam. The operative field (outer surface of crown, clamp, rubber dam, and frame) was disinfected with 5.25% sodium hypochlorite (NaOCl). The pulp chamber was accessed under manual irrigation with apyrogenic saline solution, using a sterile high-speed diamond bur. Biomechanical instrumentation was performed 1 mm short of the root canal length using the Reciproc system (VDW, Munich, Germany) adapted to an electric motor (VDW, Munich, Germany) in reciprocation movement, according to the manufacturer's protocol. The working length (WL) was determined with a Root ZX II apex locator (J Morita Corp, Kyoto, Japan) by introducing a #15 K-type hand file (Dentsply Maillefer, Ballaigues, Switzerland) up to the apical foramen, followed by radiographic confirmation. Irrigation was performed with 5 mL of 1% NaOCl using a 30-G NaviTip needle (Ultradent Products Inc, South Jordan, UT, USA), 2 mm short of the WL after each file insertion. The irrigant remained in the root canal throughout the entire procedure.

After instrumentation, the root canals were irrigated with 5 mL of 17% EDTA solution (Inodon, Porto Alegre, RS, Brazil), which remained within the root canal for 3 min without any agitation. Finally, the canals were rinsed with 10 mL of saline solution and dried with Reciproc paper points (VDW, Munich, Germany). Subsequently, the roots were filled with AH-Plus sealer (Dentsply; DeTrey, Konstanz, Germany) and single Reciproc gutta-percha cone (VDW, Munich, Germany), 1 mm short of the root canal length, and according to the manufacturer's instructions. In order to avoid a potential sealer extrusion to the periapical tissues, cement contact with the end of the gutta-percha cones was avoided. Endodontic treatments were concluded by sealing the coronal access with glass ionomer cement (SS White Goods Dental Ltd, Gloucester, England) and light-cured composite resin (Z-100, 3M, Saint Paul, USA) to avoid contamination. Then, a final periapical radiograph was obtained to confirm the adequate filling of the root canal space. The patient did not present any painful symptoms and did not need analgesics during and after treatment.

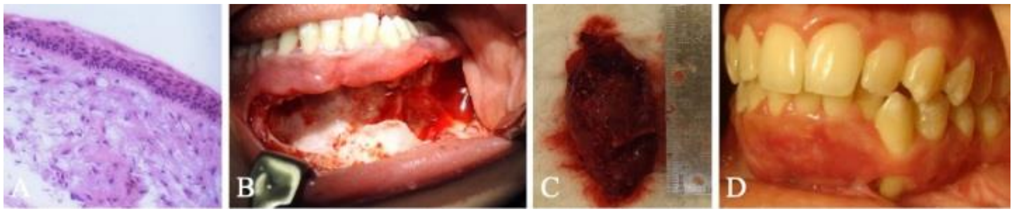

***Figure 3**. A)* Photomicrograph of OKC wall showing parakeratinised lining epithelium, displaying a palisaded basal layer with basophilic nuclei and surface corrugations (haematoxylin and eosin stain, original magnification 200×); *B)* Intraoral view after exeresis and curettage of the lesion (June 2017); *C)* Enucleated OKC with a 5-cm extension; *D)* Intraoral view one month after surgery, showing the root apices of teeth 33 to 36 exposed to the oral cavity (July 2017)

After one month (Figure 1B), teeth 33 to 36, which had root apexes exposed to the oral cavity due to the surgical removal of the lesion (Figure 3D), went through apical surgery in order to create an appropriate bone development (Figure 1C). Periodic clinical and orthopantomogram examinations were carried out one month, three months, six months, and one year after completion of the treatment. The panoramic radiographic image showed bone formation after one year of follow-up (Figure 1D). Currently, the patient presents no signs and symptoms of recurrence after one year and there are no radiopaque images suggestive of new pathology (Figure 1D). Clinical and radiographic follow-up will be maintained for at least five years, considering the mean recurrence time is 3-5 years after the initial treatments [7]. The patient signed an informed consent form, authorizing the publication of the case and any accompanying images.

Discussion

In this clinical case, an 18-year-old female patient was diagnosed with OKC in her mandible by means of histopathological examination, after imaging examinations (orthopantomogram and CBCT) showed a suspicious lesion. According to the literature, OKC is a lesion typically asymptomatic, and is usually found incidentally by means of radiographic examination [6]. It appears as well-defined unilocular or multilocular radiolucent lesions in jaws, with tendency to grow in an anterior-posterior but minimal buccolingual expansion [2, 4, 5]. Root resorption of adjacent teeth and root displacement were observed in CBCT and orthopantomogram images. However, they are occasional signs in OKC, unlike other odontogenic lesions such as ameloblastoma [5]. The present case fulfilled the histopathological criteria for OKC, which had a cystic space containing desquamated keratin with a lining of 6- to 10-cell layer parakeratinised stratified squamous epithelium, with uniform thickness and a hyperchromatic palisaded basal cell layer [4, 9].

Several treatment options for OKC have been suggested in the literature, ranging from the most conservative such as marsupialization or decompression to the most radical such as resection [1, 2, 4, 7, 9, 11, 12]. The choice of the treatment depends mainly on the size and location of the lesion, the relationships with surrounding structures, and whether OKC is primary or recurrent [11, 13]. Although marsupialization and decompression present minimal technical differences, they are considered synonyms. They both aim to relieve intra-cystic pressure through drainage to the oral cavity, allowing gradual decrease of the cystic cavity and bone apposition before OKC extirpation. Both are indicated in cases of large intra-osseous cysts, with risk of damaging either important anatomical structures or pulp vitality [2, 11, 13]. Alternatively, enucleation is performed when OKC is not large, and when only minor damage to adjacent tissues is expected, as observed in the present case, considering that the teeth involved in the lesion already presented pulp necrosis [13]. Finally, aggressive large lesions in adults or recurrent OKCs should be treated by resection [2, 7, 9, 12].

Previous studies have also focused on the relationship between surgical management and recurrence rate of OKCs [1, 7, 12, 14]. These studies suggest that the recurrence of the lesion is mainly a consequence of operative factors. Recurrence of OKCs may be due to the following factors: incomplete removal of the original cyst, growth of new OKC from odontogenic epithelial remnants or from satellite cysts left in the surgical wound, or development of a new lesion in adjacent tissues interpreted as a recurrence [15]. Although resection is the surgical method with the lowest recurrence rates [1, 7, 12], enucleation is currently the most used treatment of OKC, due to the high morbidity reported after resection and resistance of the patients to accept a more radical treatment of a benign lesion [7, 12]. However, in order to reduce the risk of recurrence, adjuvant surgical methods such as curettage, chemical cauterisation with Carnoy's solution, liquid nitrogen cryotherapy, or peripheral ostectomy are indicated after enucleation [4, 7, 14].

Pulp necrosis occurs mainly due to an inflammatory process triggered by infection and it may be accompanied by the development of a periapical lesion (periapical granuloma, periapical abscess, or periapical cyst) [16, 17]. However, a wide variety of lesions may cause aseptic pulp necrosis and mimic an endodontic periapical lesion, especially OKC, making diagnosis and treatment choice difficult [3, 8]. Thus, although oral and maxillofacial surgeons receive most cases of non-endodontic lesions [8], endodontists and general dentists should understand the characteristics of this pathology and the treatment options for the lesion and other damaged anatomical structures, in order to prevent an incorrect diagnosis and elaborate an adequate multidisciplinary treatment plan. In the present case, due to the possibility of maintaining the natural teeth after surgically removing the lesion, the patient underwent endodontic treatment of teeth 36 to 45 and apicoectomy of teeth 33 to 36.

Considering that all teeth presented aseptic pulp necrosis without bacterial involvement with the aetiology of OKC, the endodontic treatment was performed in a single session to reduce both the risk of contamination between sessions and treatment time [18]. In addition, several studies have shown no difference in the quality of root canal treatment and in the repair of periapical lesions when teeth are treated in a single session or in multiple sessions using intracanal dressing between them [18-21]. Paredes-Vieyra *et al.* [21] performed a randomized controlled trial including 300 non-vital teeth with apical periodontitis and classified 96.57% of teeth treated in a single session as healed compared with 88.97% of teeth treated in two sessions. Thus, single-session endodontic treatment may be as effective as the multi-session treatment when biomechanical preparation is adequate [21].

The working length during instrumentation was defined at 1 mm from the root apex, because the pulp cavity was aseptic and there was no need for mechanical cleaning of the cemental canal [22]. This case aimed to remove the necrotic tissue and shape the dentinal root canal. Mechanical instrumentation of the root canals was performed using the reciprocating single-file (Reciproc) technique, because it allows better control of apically extruded dentin and pulp debris in relation to conventional hand-file and multi-file rotary systems [23-26]. The instrumentation technique was a concerning factor in the endodontic approach of this clinical case, because apical extrusion of debris and irrigating solutions could induce inflammation, postoperative pain, and delay of surgical wound repair [23, 25, 26].

Conclusion

Endodontic treatment associated with the surgical enucleation and posterior curettage of odontogenic keratocyst (OKC) contributed positively to the outcome of the clinical case because the patient showed significant mandibular bone neoformation and no signs of recurrence after one year of follow-up. The multidisciplinary treatment protocol presented may be a good alternative to preserve anatomical structures and teeth in cases of extensive and multilocular OKC, maintaining the quality of life of patients.

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Conflict of Interest: 'None declared'.

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