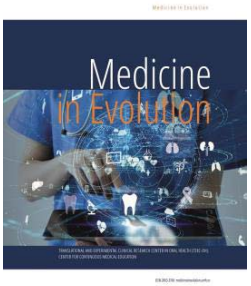
A Case of Odontogenic Keratocyst in the Posterior Ramus and Body of the Mandible



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Abstract

The dental lamina gives rise to the odontogenic keratocyst (OKC), a common odontogenic cyst that affects the maxillofacial region. The OKC is distinct from other jaw cysts and is more likely to return when combined with aggressive clinical behaviour. The recurrence rate in OKC is 25%–30%. In 2005, the World Health Organization (WHO) group categorized odontogenic keratocyst (OKC) as a tumour and proposed the abbreviation KCOT to differentiate the condition from the ortho keratinizing variant. The WHO reclassified KCOT as OKC in 2017 based on data demonstrating non-neoplastic clinical behaviour. The 19-year-old male in this case study has OKC in the ramus and body of his jaw, near the left mandibular molar tooth. This case study aims to demonstrate the need for dentists to do in-depth investigations into each circumstance and offer patients better treatment options with ongoing patient monitoring and follow-up. Enucleation, marsupialization, and other surgical techniques are possible; however, in this case, our objective was to preserve the patient's mandible and facial features.

Keywords: Keratocyst, odontogenic cyst, mandible

INTRODUCTION

The cystic lesions that most frequently affects the maxillofacial region are odontogenic cysts. They are generally divided into two groups: one for development, which includes keratocyst and dentigerous cysts, and the other for inflammation, which includes radicular cysts [1]. The odontogenic keratocyst (OKC) was first described by Philipsen (1956), in 2005, it was designated by the World Health Organization as a keratocystic odontogenic tumor (KCOT). Then reclassified as odontogenic keratocyst (OKC) in 2017 [2]. It is "a benign unicystic or multicystic intraosseous tumor of odontogenic origin, with a unique lining of para keratinized stratified squamous epithelium and propensity for aggressive, infiltrative tendencies" [3]. The percentage of OKC versus other cysts of the jaws as given by different authors: Hjorting-Hansen et al. and Toller 11%; Brannon and Payne 9%; and Pindborg and Hansen 7% [4-9]. The lesions can grow remarkably without appreciably deforming the jaw skeleton because growth occurs mostly in the antero-posterior dimension. The propensity for rapid growth is brought on by increased osteolytic activity of prostaglandin substances in the cell population of the cyst lining, higher activity of the epithelial cells lining the cyst, and higher accumulation of hyperkeratotic scales in the cyst lumen, resulting in a greater hydrostatic pressure difference [10]. The OKC has a unique and prevalent clinical and histologic lesion with aggressive nature. It often develops in the dental lamina, but some speculate that basal cell component may be its likely source [11]. The etiology of KCOT is related to the development of the dental lamina and, in particular, remnants of it after it has served its purpose. These dental lamina-derived epithelial islands are primarily seen in the periodontal ligament and gingiva. It clarifies the clinical entity of OKCs lateral follicular or periodontal appearance [12]. The characteristic features of OKC are the tendency to grow along the cancellous channels with very little cortical expansion. Numerous hypotheses regarding the growth of KCOT have been put forth. These include intraluminal hyperosmolality, active epithelial proliferation, the collagenolytic activity of the cyst wall, and synthesis of interleukin 1 and 6 by keratinocytes which will induce the secretion of keratocyte growth factor from interactive fibroblasts along with tumor necrosis factor leading to increased levels of prostaglandins and expression of the parathyroid related protein [12]. The mandible is involved in 70% or more cases, particularly in the third molar, angle, and ramus regions. Next, the most common site of occurrence is the maxillary third molar, followed by the mandibular premolar.

MATERIAL AND METHODS

A 19-year-old male patient reported to our dental institute with mild pain and swelling in the lower left backregion of the mandible. The patient was sent from a dental private practice after the dentist saw the orthopantomography (Figure 1).

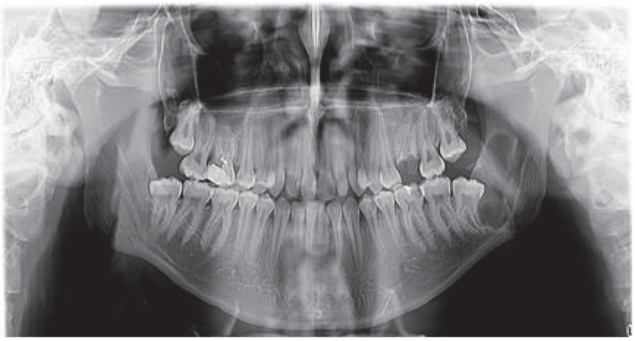

Figure 1. Unilocular radiolucency with scalloped and corticated margins seen in the mandibular posterior left body and ramus extending next to the tooth 38

An unilocular radiolucency can be seen in the mandibular posterior left of the body. Computed tomography was done to examine the extent of the lesion three-dimensionally (Figure 2).

Figure 2. Sagital, coronal and axial images of the CT revealed an unilocular radiolucency

The computed tomography revealed in the left mandibular angle, next to the root of the tooth 38 a well-defined unilocular cystic mass, with axial diameter of approximately 7/11 mm and cranio-caudal diameter of 30 mm, which thins the cortical bone. Antibiotic and anti-inflammatory therapy was started.

After taking the informed consent the patient was scheduled for surgery (Figure 3) In analgesia, local anesthesia was performed, standard triangular flap (Figure 4), cyst enucleation with curettage (Figure 5), histopathological examination, tooth extraction 38 (Figure 6), wound closure.

Figure 3. Prepared sterile surgical instruments

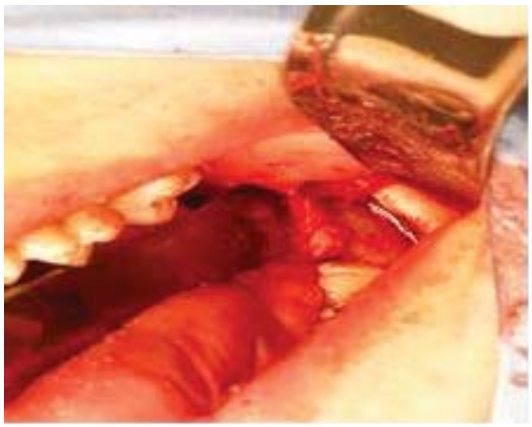

Figure 4. Intraoral view after cyst extirpation

Figure 5. Excisional biopsy of the cyst

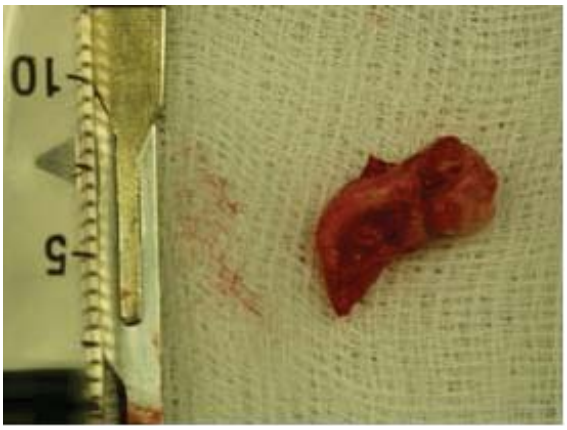

Figure 6. Tooth 38 after surgical extraction

The excisional biopsy revealed cell layer thick para keratinized stratified squamous epithelium and necrotic epithelium.

RESULTS

The evolution of the patient was favourable, he respected the indications and the compliance was very good. Preoperative and postoperative antibiotics and anti-inflammatory regimen was followed. The suture was removed after 8 days. The postoperative course was uneventful and the patient is still being followed-up (Figure 7).

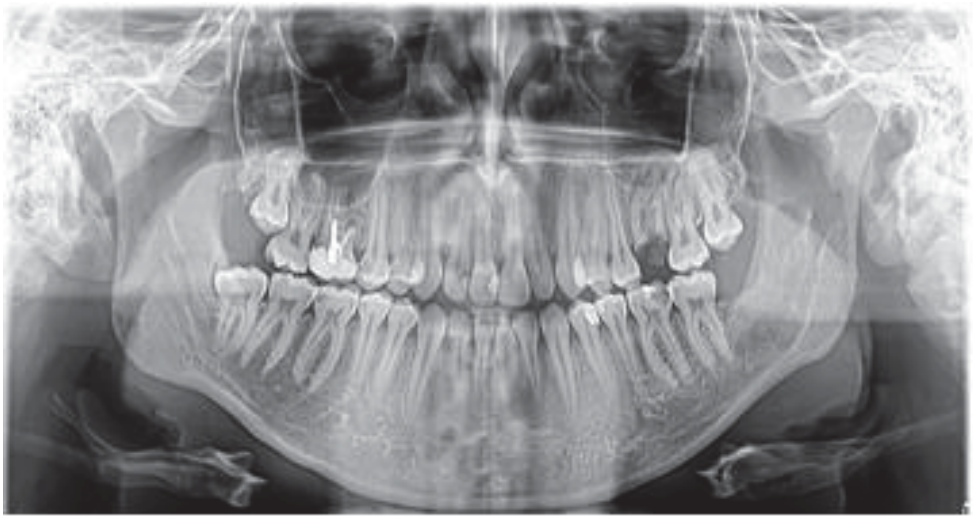

Figure 7. Orthopantomography 7 months after surgery

DISCUSSIONS

According to a study conducted by Borgehesi A et al. in 2018, OKC account for around 10% of all odontogenic cysts. The reported age distribution ranges from 8 to 82 years, with a high occurrence in the third decade of life. There is a slight male bias [13]. The same findings were observed in our case. A study by Hasen EH et al. in 1969 highlighted that keratocyst in the jaw appeared to be non-inflammatory. The epithelium is most likely derived from epithelial remnants from the dental lamina, and the researchers concluded that the dental lamina could yield keratinized epithelium [5]. Toller PA et al. in 1972 featured that the

keratinizing epithelium surrounding the odontogenic keratocyst shows cell maturation rather than degeneration and can expand in size primarily through epithelial cell multiplication. The turnover of epithelial cells in odontogenic keratocyst is higher than in other cysts [6]. Yazdani J et al. in 2009 pointed out that OKC is unique among jaw cysts as it frequently recurs and is related to aggressive clinical behavior. OKC has a risk of relapse of 25-30% [1]. Brannon RB and Pinborg JJ suggested in their study that OKC has a peak incidence in the second and third decades of life. The mandible to the maxilla ratio was 2:1, and the ramus and third molar regions of the jaw were the most often used areas. The same was observed in this case; the lesion involved the mandible's body up to the mandible's angle and ramus. The dentigerous cyst was the most common diagnosis for keratocyst. These cysts may be very aggressive clinically. Their radiographic appearance was quite variable, although they resembled ameloblastomas; the same appearance was evident in our case, which caused a dilemma in arriving at a diagnosis. They have a high recurrence rate compared to other types of odontogenic cysts [7,9].

CONCLUSIONS

In this case report, a 19-year-old male patient with a cystic swelling clinically and radiographically mimicking an ameloblastoma in the left side ramus and body of the mandible. The histopathologic report supported the diagnosis of OKC. This OKC was discovered accidentally by a routine investigation at the dentist after a routine orthopantomography. The affected tooth, 38 was extracted, the cyst was enucleated completely and a long-term follow-up was conducted. It showed proper healing (Figure 9), and as of this writing, OKC has not returned. Enucleation, marsupialization, and other surgical techniques are possible; however, we aimed to preserve the patient's mandible and facial features in this case. This case study intends to demonstrate the need for dentists to thoroughly evaluate each scenario and provide better treatment options with long-term patient follow-up.

REFERENCES

1. Yazdani J, Kahnamouii SS. Developmental odontogenic cysts of jaws: a clinical study of 245 cases. J Dent Res Dent Clin Dent Prospects. 2009 Spring;3(2):64-6.
2. Soluk-Tekkeşin M, Wright JM. The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2017 (4th) Edition. Turk Patoloji Derg. 2018;34(1):10.5146/tjpath.2017.01410.
3. Madras J, Lapointe H. Keratocystic odontogenic tumor: reclassification of the odontogenic keratocyst from cystto the tumor. J Can Dent Assoc. 2008 Mar1;74(2):165a-h.
4. Langland OE, Langlais RP. Diagnostic Imaging of Jaws: focal radiopacities of jaw. 2nd ed. Baltimore: Williamsand Wilkens; 1995.505-15.
5. Hjorting-Hansen E, Andreasen JO, Robinson LH. Astudy of odontogenic cysts, with special reference tolocation of keratocysts. Br J Oral Surg. 1969 Jan1;7(1):15-23.
6. Toller PA. Newer concepts of odontogenic cysts. Int JOral Surg. 1972;1(1):3-16.
7. Brannon RB. The odontogenic keratocyst. A clinicopathologic study of 312 cases. Part I. Clinical features. Oral Surg Oral Med Oral Pathol. 1976 Jul1;42(1):54-72.
8. Payne TF. An analysis of the clinical and histopathologic parameters of the odontogenic keratocyst. Oral SurgOral Med Oral Pathol. 1972 Apr 1;33(4):538-46.
9. Pindborg JJ, Hansen J. Studies on odontogenic cyst epithelium: 2. Clinical and Roentgenologic Aspects of Odontogenic Keratocysts. Acta Patho Microbiol Scand. 1963 Sep;58(3):283-94.
10. Haring JI, Van Dis ML. Odontogenic keratocysts: aclinical, radiographic, and histopathologic study. OralSurg Oral Med Oral Pathol. 1988 Jul 1;66(1):145-53.

11. Soni PK, Choudhary B, Vashistha V, Sinha P. Odontogenic keratocyst in posterior mandible: A case report. Int J Sci Study. 2021 Aug 30;9(1):4-7.
12. Menon S. Keratocystic Odontogenic Tumours: Etiology, Pathogenesis and Treatment Revisited. JMaxillofac Oral Surg. 2015 Sep;14(3):541-7.
13. 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 Oct;9(5):883-97.