

Odontogenic keratocyst in the mandible, surgical treatment and reconstruction: A case report

Ezgi Aydın, Neşe Kurt Özkaya, Halit Şengel

ABSTRACT

Introduction: Odontogenic keratocyst is a locally aggressive and highly recurrent lesion, which occurs mostly on the posterior mandible with a slightly higher rate in males. It is thought to originate from dental laminae residues. Radiographically, it may appear unilocular or multilocular, with or without defined borders. **Case Report:** In this case report, surgical treatment of the odontogenic keratocyst in the right posterior mandible and reconstruction of the defected site is presented. **Conclusion:** Due to highly recurrent nature of the odontogenic keratocysts, patient's follow-up continues.

Keywords: Dental implants, Enucleation, Extraoral bone graft, Odontogenic keratocyst, Reconstruction

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INTRODUCTION

Odontogenic keratocyst was first described by Philipsen in 1956 and has been a subject to many researches over the years. The aggressive nature and high recurrence rate of the lesion makes it difficult to treat the lesion [1].

Odontogenic keratocysts are considered to originate from the dental laminae residues. They occur predominantly between 10 and 30 years of age, usually in the posterior mandible, accounting for approximately 12–14% of all cystic lesions. The incidence rate in males is slightly higher in respect to females. Radiographically, the lesion may show both unilocular and multilocular radiolucency [2].

Although there are various documented treatments for the odontogenic keratocyst in literature, there is no universally accepted treatment approach. Treatment options include enucleation (with or without curettage), decompression and marsupialization, peripheral osteotomy, cryotherapy, application of Carnoy's solution and resection of the jaw [2].

Odontogenic keratocyst was named a keratocystic odontogenic tumor by the World Health Organization (WHO) in 2005 due to its local aggressive nature and high recurrence rate, budding in the basal layer, the appearance of mitotic figures in the suprabasal layer and the mutation of PTCH gene in the syndromic lesions [3].

CASE REPORT

A 25-year-old male was referred to our clinic with a complaint of pain in the posterior mandible. He had good

oral hygiene and a history of odontogenic keratocyst surgery. Radiographical examination of the patient revealed a radiolucency between the right 2nd premolar and the 1st molar teeth with well-defined borders extending from the coronal of the alveolar bone to below the apexes of the adjacent teeth and not reaching the mandibular nerve (Figure 1).

There were no former radiographs of the patient but patient's history of having been operated for odontogenic keratocyst in the identical area led us to think that it is a recurrence of the former keratocyst. The patient had no medical problems and it was decided to perform enucleation and curettage with the preliminary diagnosis of recurrent odontogenic keratocyst. Surgical site was reached with sulcular and vertical incisions under local anesthesia using 2 ml (one tube) of maxicaine fort (articaine hydrochloride 40 mg/ml, epinephrine hydrochloride 0.012 mg/ml, Vem Ilac; Turkey) with mandibular technique. Upon observing that the tooth roots were related to the lesion, lower right 2nd premolar and 1st molar teeth were extracted. The entire lesion was removed and curettage was performed. The operation area was closed with 3-0 silk suture (Jinhuan Medical Products LTD/China). The patient was prescribed 1000 mg amoxicillin and clavulanic acid (GlaxoSmithKline) two times daily, 550 mg naproxen sodium (Abdi İbrahim) three times daily and chlorhexidine gluconate (Pharmactive) two times daily for a week. The histopathologic examination was concluded with the diagnosis of odontogenic keratocyst.

While the patient was under postoperative control, a radiolucent lesion distal to the right lower third molar tooth with well-defined borders was observed and the removal of the lesion with the extraction of adjoining tooth was planned (Figure 2).

Surgical site was reached with sulcular and vertical incisions under local anesthesia using 2 ml (one tube) of maxicaine fort (articaine hydrochloride 40 mg/ml, epinephrine hydrochloride 0.012 mg/ml, Vem Ilac, Turkey) with mandibular technique. Lower right 3rd molar was extracted and the lesion was removed and issued for pathological examination. The operation area was closed with 3-0 silk suture (Jinhuan Medical Products LTD/China). The patient was prescribed 1000 mg amoxicillin and clavulanic acid (GlaxoSmithKline) two times daily, 550 mg naproxen sodium (Abdi İbrahim) three times daily and chlorhexidine gluconate (Pharmactive) two times daily for a week.

Once the histopathologic examination revealed the lesion to be an odontogenic keratocyst, the patient was consulted for the evaluation for the Gorlin (Nevoid Basal Cell Carcinoma) syndrome. The PTCH1 gene mutations were examined and the patient was not found to have a genetic mutation.

Following nine months of follow-up and ensuring that there was adequate recovery of the soft tissue and bone in the area with no evidence of recurrence, reconstruction

of the area with implants and extraoral autogenous bone grafts was planned (Figure 3–4).

Patient was consulted with plastic reconstructive and aesthetic surgery department and was decided to be operated simultaneously under general anesthesia for harvesting extraoral autogenous bone grafts from the iliac bone and the reconstruction of the defected area. Horizontal and vertical incisions were performed to reveal the defect (Figure 5).

Under sufficient irrigation, the slots were prepared using the burs according to the manufacturer's recommendation. The implants were placed and the defect was filled with the autogenous graft from the iliac bone (Figure 6).

The operation area was closed with 3-0 silk suture. The patient was prescribed 1000 mg amoxicillin and

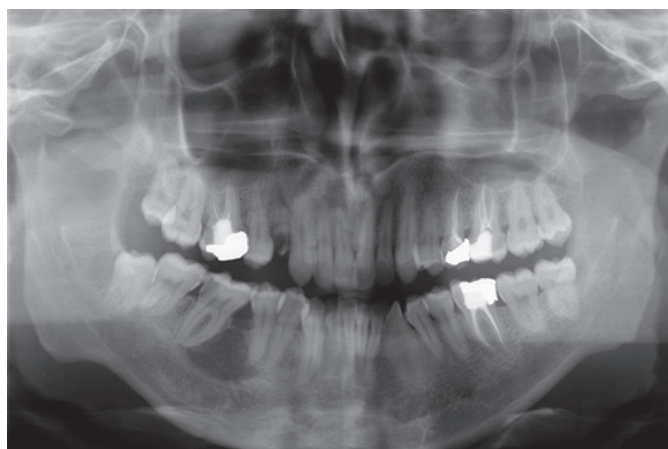


Figure 1: Initial radiographic evaluation of the patient.



Figure 2: Healing after the removal of the cyst and the extraction of the adjacent teeth, lesion related to lower right 3rd molar.



Figure 3: Follow-up after enucleation of cysts (Postoperative 9th month).

clavulanic acid (GlaxoSmithKline) two times daily, 550 mg naproxen sodium (Abdi İbrahim) three times daily and chlorhexidine gluconate (Pharmactive) two times daily.

After approximately six months of follow-up, healing caps were placed on the implants. There was no problem with healing or osseointegration (Figure 7).



Figure 7: Monitoring of osseointegration after six months from reconstruction.

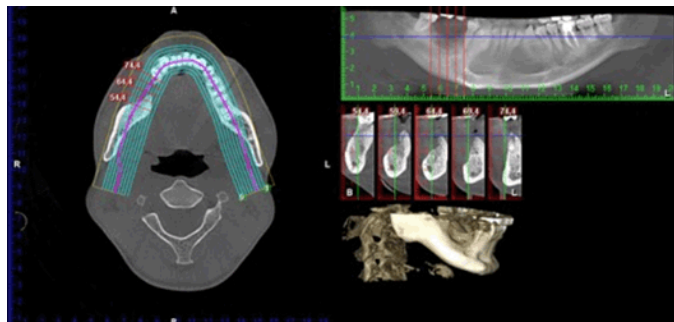


Figure 4: Postoperative ninth month tomography image.

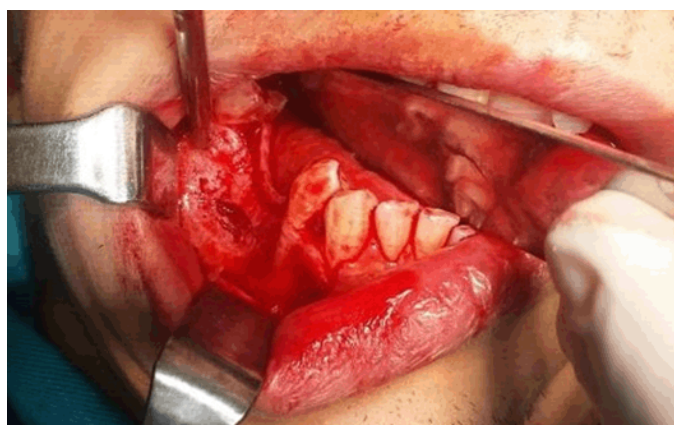


Figure 5: Appearance of the defect in the enucleated area.



Figure 6: Defected area, grafted and implanted.

DISCUSSION

The correct diagnosis and treatment of odontogenic keratocysts is crucial for three reasons:

- Keratocysts are considered more aggressive than other odontogenic cysts [4].
- Its recurrence rate is higher than odontogenic cysts [5].
- The probability of being associated with neoplastic basal cell carcinoma syndrome makes the patient with multiple cysts a candidate for the syndrome [6–8].

Odontogenic keratocysts can be seen in a wide range from childhood to old age. However, 60% of the cases are reported between 10–40 years of age [9]. The incidences in males is slightly higher than that of females, and 60–80% are seen especially in the posterior mandible [10].

In this report, a 25-year-old male patient with an odontogenic keratocyst in the posterior mandible was observed in accordance with the common form of the cyst.

In various studies odontogenic keratocysts ranged from 2.5–62.5% for recurrence [5]. Regezi et al. reported a recurrence rate of 10–30% for single odontogenic keratocysts and that multiple sporadic jaw cysts were seen without syndromes in approximately 5% of patients with odontogenic keratocysts with a higher recurrence rate than that of single lesions [11].

Nevoid basal cell carcinoma syndrome (Gorlin syndrome) is an autosomal dominant disease caused by mutation in PTCH tumor suppressor gene. Patients may have temporoparietal bossing, hypertelorism, mandibular prognathism and skeletal anomalies [8]. The most prominent clinical feature is the predisposition to develop multiple basal cell carcinomas. The appearance of multiple odontogenic keratocysts is usually the first manifestation of the syndrome. Therefore, any patient with odontogenic keratocysts should be evaluated for this syndrome.

Williams and Connor recommended primer enucleation and curettage for the treatment of odontogenic keratocysts and recommended the application of Carnoy's

solution (composed of 60% ethanol, 30% chloroform and 10 % glacial acetic acid) for three minutes following methylene blue as a marker agent [5]. In the present case, Carnoy's Solution was not used as the ingredients of the solution forms a risk for paresthesia if used near the mandibular nerve. In the case of recurrence, they reported that the resection should be performed with appropriate bone and soft tissue borders. MacIntosh argues that resection of odontogenic keratocysts with a 5 mm linear margins should be the primary treatment modality [12]. However, resection or curettage made with wide margins makes it difficult to reconstruct the defect afterwards.

There are several options for harvesting bone graft from extraoral donor sites. Between the iliac crest, costae, fibula, radius, tibia and calvarium, the iliac crest was chosen due to being most commonly used donor site for oral reconstructions, easier to operate with high volume of graft available and easily accepted by the patient.

CONCLUSION

Multiple odontogenic keratocysts in our patient required us to choose an extraoral donor site for the graft. There is no general consensus as to how long the odontogenic keratocysts should be followed for recurrence. There is no indication of recurrence during this follow-up period. The patient's follow-up is going on.

Author Contributions

Ezgi Aydın – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Neşe Kurt Özkaya – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Halit Şengel – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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