

Resection and Immediate Reconstruction of a Large Mandibular Keratocystic Odontogenic Tumor: A Case Report

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ABSTRACT

Keratocystic Odontogenic Tumor (KCOT) is a benign yet aggressive cystic lesion of the jaws, often associated with high recurrence rates and locally destructive behavior. It accounts for 10%–20% of all odontogenic cysts, commonly affecting the mandible and presenting in the second to fourth decades of life. Although the World Health Organization initially classified KCOT as a cystic neoplasm due to its aggressive nature and genetic mutations, it was later reclassified as an odontogenic cyst following evidence that partial treatment may lead to regression. Treatment options for KCOT vary, with resection favored for larger, more aggressive lesions due to high recurrence rates with conservative approaches. This case report presents a 38-year-old male with recurrent jaw pain and locking due to a large KCOT in the right mandibular condyle. The patient underwent resection and immediate reconstruction using a custom-made total joint prosthesis. Post-operative recovery and follow-up are discussed, along with relevant imaging findings.

Key words: Keratocystic Odontogenic Tumor

Odontogenic Cysts

Mandibular Condyle

Arthroplasty, Replacement

Reconstruction, Surgical

INTRODUCTION

Keratocystic Odontogenic Tumor (KCOT) is a benign yet aggressive developmental cystic lesion of the jaws that originates from the remnants of the dental lamina and may occur sporadically or in association with nevoid basal cell carcinoma syndrome (NBCCS) (1). KCOT accounts for 10%–20% of all odontogenic cysts, ranking as the third most common cystic lesion in the jaws after radicular and dentigerous cysts (2). Due to its aggressive behavior and high recurrence rates (up to 62.5%), it has been debated whether it should be

considered a cyst or a neoplasm (3). In 2005, the World Health Organization (WHO) initially classified KCOT as a cystic neoplasm due to its association with PTCH gene mutations (3). However, in 2017, WHO reclassified KCOT as an odontogenic cyst, following new evidence that such lesions may regress after partial treatment, contradicting the behavior expected of a neoplasm (4).

KCOTs commonly occur in the mandible, with peak incidence in the second to fourth decades of life (5). Management options range from marsupialization and decompression to resection. The best treatment approach remains debated due to high recurrence rates associated with conservative treatments (1, 6). The article presents a report of a case with KCOT, which underwent resection and reconstruction.

Case Presentation

A 38-year-old male from Isfahan presented to the clinic on 4 March 2024 with complaints of periodical swelling in the right parotid area and recurrent locking of the jaw. The patient reported that the jaw pain and intermittent episodes of "closed lock" started approximately two years ago. The initial episode occurred without any obvious trauma, and he experienced a sudden inability to fully open his mouth. However, the patient had no signs of pain or limitation of mouth opening during examination. Neurological exam of the facial nerve was normal. (Fig. 1)



Figure 1. The patient had no signs of pain or limitation of mouth opening during examination.

An orthopantomogram (OPG) performed one year ago revealed an impacted third molar, which was subsequently extracted by a dentist. Despite the procedure, the patient continued to experience intermittent episodes of jaw locking and associated pain over the next six months. He was hospitalized for one week during this period and treated with antibiotics, analgesics, and adjunctive physical therapy. Despite this intervention, his symptoms persisted.

Following continued discomfort, the patient consulted with an ENT specialist, who initiated another course of antibiotic therapy, along with dexamethasone and analgesics. However, the symptoms persisted. A CT

scan was performed (Fig 2), and two cystic lesions (2×2 cm) were observed in the right mandibular, extended from condyle to ascending ramus. Based on the findings, a differential diagnosis of Keratocystic Odontogenic Tumor and ameloblastoma was made. Thus, he was referred to a maxillofacial surgeon for further evaluation and management.

Due to the large size of cystic lesions, resection and reconstruction were chosen in one step. A virtual surgery was performed for the patient, with the lesion defined at a safe margin of 1 centimeter. A stent and a cutting guide were meticulously designed for the patient, followed by the creation of custom-made total joint prostheses (Fig. 3). On the day of the surgery, after accessing the lesion through a submandibular and retromandibular incisions, the lesion was carefully resected by using the cutting guide (Fig4), and the total joint prosthesis was then placed in position and patient's occlusion was assessed and confirmed during surgery. (Fig. 5)

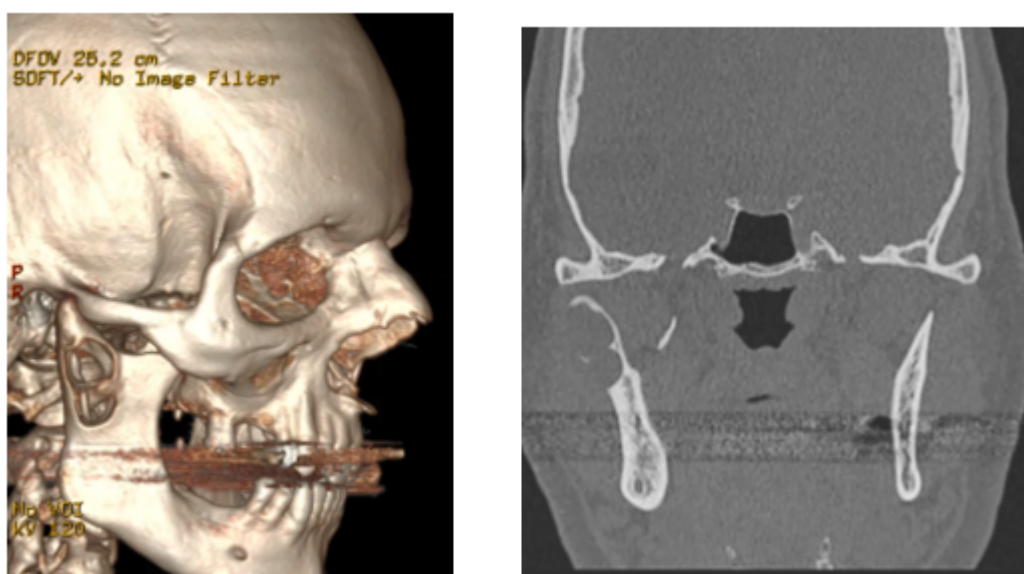


Figure 2. Pre-operative CT scan with 3D reconstruction (left) and coronal view (right)

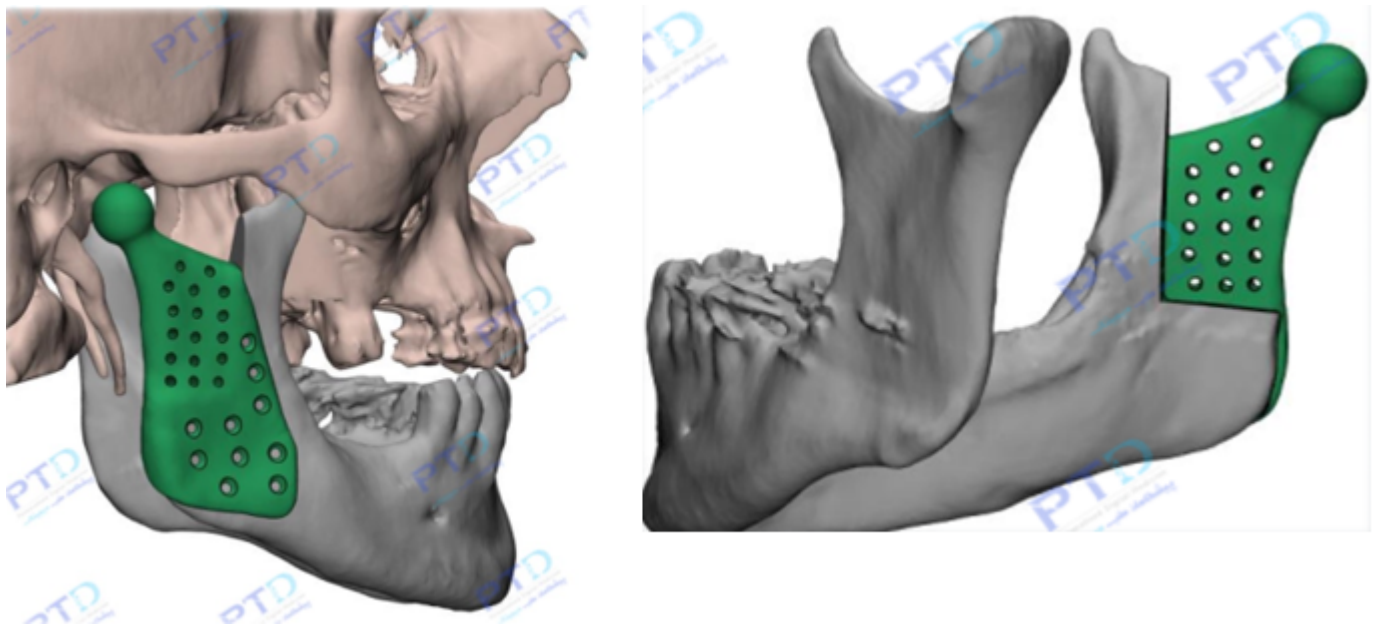


Figure 3. 3D reconstructed plate with an attached metal joint



Figure 4. Cutting Guide using for precise resection(Left) Resected part of right mandible, two cystic lesions are visible(Right)



Figure 5. Patient's occlusion was assessed and confirmed during surgery.

Histopathological examination revealed fibrotic tissue infiltrated by chronic inflammatory cells and ulcerative epithelium with palisaded hyperchromatic basal cells. Thus, the lesion was diagnosed as a *Keratocystic Odontogenic Tumor* involving the right mandibular condyle.

At one-year follow-up, the patient's mouth opened adequately, and all neurological assessments of the facial nerve were normal, with no deviations in the patient's jaw (Fig. 6). CT scan shows no new pathological lesions or structural defects of the prosthetic joint after one year. (Fig. 7)



Figure 6. Resolution of patient symptom and ability to fully open mouth at one-year follow up(Left)
Functional facial nerve one year after surgery (Right)

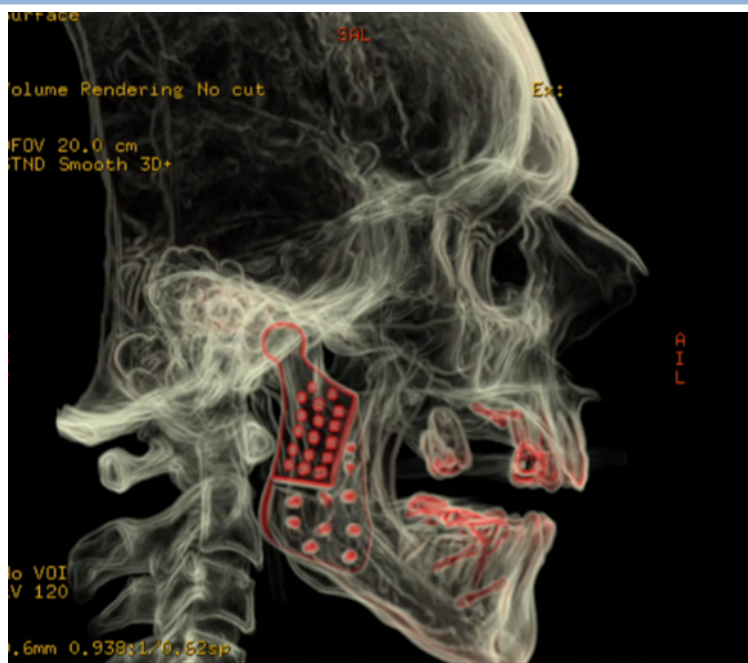


Figure 7. CT scan after one year shows no new pathological lesions or structural defects of the prosthetic joint

Discussion

Keratocystic Odontogenic Tumors (KCOTs) are aggressive, recurrent lesions of the jaw, often requiring a tailored surgical approach to achieve optimal outcomes (1). In this case, the patient presented with a right mandibular KCOT involving the condyle, for which radical resection was performed following persistent symptoms and previous unsuccessful conservative treatments. The differential diagnosis for lesions such as this includes ameloblastoma and KCOT. These lesions can exhibit similar clinical and radiographic features, necessitating histopathological evaluation to confirm the diagnosis.

Treatment modalities for KCOTs have been extensively reviewed in the literature. Herb (2006) provided a comprehensive overview of the different treatment options (7), while Ghali and Connor focused on the surgical management of keratocystic odontogenic tumors (KCOTs), a term previously used for KCOTs (8). Treatment strategies for KCOTs include enucleation and curettage, enucleation with peripheral ostectomy, complete resection, cryotherapy, chemical adjunct therapy, decompression, and marsupialization. Of these, enucleation and curettage, although conservative, are associated with the highest recurrence rate and are no longer widely recommended as the initial treatment for KCOTs (1). Peripheral ostectomy is often considered an adjunct to less radical treatments, but as Ghali and Connor noted, determining the appropriate depth of bone removal remains a challenge. Methylene blue dye has been suggested as a method to stain the cyst periphery, but its efficacy is uncertain due to the unknown penetration depth (8).

In this case, radical resection was chosen due to the size, aggressive nature, and condyle involvement. As Ghali and Connor highlighted, osseous resection is one of the most extensive treatments. However, it offers a higher success rate as the lesion is removed en bloc with a margin of surrounding bone, typically 1 cm. This approach is particularly effective when cortical perforation or soft tissue involvement, as seen in our patient (8). If indicated by bony dehiscence, soft tissue resection can further reduce recurrence rates.

Other adjunctive treatments, such as cryotherapy and the use of Carnoy's solution, have been explored to

manage residual cystic cells after surgical removal. Cryotherapy, by freezing the lesion site, helps to destroy any remaining cystic cells, reducing recurrence. Similarly, chemical cauterization using Carnoy's solution has been shown to lower recurrence rates, although it may pose risks to surrounding tissues (9).

Decompression and marsupialization are additional conservative treatment options, often reserved for large or multilocular cysts (7). Marsupialization can be a one-stage definitive treatment, while decompression is typically a two-stage procedure involving the deroofting of the cyst followed by cystectomy after several months. While these options may reduce the size of the lesion and avoid the need for more aggressive surgery, they are generally not recommended as standalone treatments for aggressive or recurrent KCOTs (8).

In large, multilocular lesions, particularly those involving critical structures like the ramus or maxilla, resection may be the only viable option. In such cases, procedures like hemimandibulectomy or hemimaxillectomy may be required, followed by immediate or delayed reconstruction to restore function and aesthetics. Our patient underwent immediate reconstruction with a reconstruction plate, bone graft, and plasma-rich platelets to re-establish mandibular continuity, preventing long-term complications associated with untreated bone defects.

In conclusion, while KCOTs can often be managed conservatively in smaller lesions, large, aggressive, or recurrent cases necessitate more radical interventions like resection. Adjunctive therapies such as cryotherapy, Carnoy's solution, and peripheral ostectomy can aid in reducing recurrence, but long-term follow-up remains essential. The chosen treatment must be individualized based on the lesion's size, location, and behavior, with radical resection being a valid option for extensive lesions involving critical structures.

References

1. Fidele NB, Yueyu Z, Zhao Y, Tianfu W, Liu J, Sun Y, et al. Recurrence of odontogenic keratocysts and possible prognostic factors: Review of 455 patients. *Med Oral Patol Oral Cir Bucal*. 2019;24(4):e491-e501.
2. Aquilanti L, Mascitti M, Togni L, Rubini C, Nori A, Tesei A, et al. Non-neoplastic jaw cysts: a 30-year epidemiological study of 2150 cases in the Italian population. *Br J Oral Maxillofac Surg*. 2021;59(2):168-73.
3. Kuroyanagi N, Sakuma H, Miyabe S, Machida J, Kaetsu A, Yokoi M, et al. Prognostic factors for keratocystic odontogenic tumor (odontogenic keratocyst): analysis of clinico-pathologic and immunohistochemical findings in cysts treated by enucleation. *J Oral Pathol Med*. 2009;38(4):386-92.
4. Laino L, Russo D, Ciccì M, D'Amico C, Fiorillo L, Cervino G. Surgical conservative approach of odontogenic keratocyst tumor of the jaws. *Minerva Dent Oral Sci*. 2021;70(1):26-31.
5. Kaczmarzyk T, Kisielowski K, Koszowski R, Rynkiewicz M, Gawełek E, Babiuch K, et al. Investigation of clinicopathological parameters and expression of COX-2, bcl-2, PCNA, and p53 in primary and recurrent sporadic odontogenic keratocysts. *Clin Oral Investig*. 2018;22(9):3097-106.
6. Titinchi F. Protocol for management of odontogenic keratocysts considering recurrence according to treatment methods. *J Korean Assoc Oral Maxillofac Surg*. 2020;46(5):358-60.
7. Giuliani M, Grossi GB, Lajolo C, Bisceglia M, Herb KE. Conservative Management of a Large Odontogenic Keratocyst: Report of a Case and Review of the Literature. *Journal of Oral and Maxillofacial Surgery*. 2006;64(2):308-16.
8. Ghali G, Connor MS. Surgical management of the odontogenic keratocyst. *Oral and Maxillofacial Surgery Clinics*. 2003;15(3):383-92.
9. Gilveti C, Mahendran K, Gulati A, Barrett AW. Recurrence rate of the odontogenic keratocysts patient cohort in a UK regional maxillofacial surgery unit. *Oral Surgery*. n/a(n/a).

