**SAS Journal of Surgery**
Abbreviated Key Title: SAS J Surg
ISSN 2454-5104
Journal homepage: https://www.saspublishers.com

OPEN ACCESS

**Surgery**

# Giant Mandibular Odontogenic Keratocyst: A Case Report and Literature Review

K. Mezzat[1]*, B. Bendali[1], Msk. Hattab[1], Y. Bennaoui[1], Z. Aziz[1], M. El Bouihi[1], N. Mansouri El Hattab[1]

[1]Department of Maxillofacial, Aesthetic and Oral Surgery, Ibn Tofail Hospital, Mohammed VI University Hospital, Marrakech, Morocco

**DOI:** https://doi.org/10.36347/sasjs.2026.v12i05.033 | **Received:** 11.04.2026 | **Accepted:** 20.05.2026 | **Published:** 26.05.2026

***Corresponding author:** K. Mezzat
Department of Maxillofacial, Aesthetic and Oral Surgery, Ibn Tofail Hospital, Mohammed VI University Hospital, Marrakech, Morocco

**Abstract** **Case Report**

Odontogenic keratocyst is a benign odontogenic cystic lesion whose biological behavior contrasts with its non-malignant histological nature. This lesion is known for its capacity for intraosseous extension, its local destructive potential, and its marked tendency to recur. Giant forms remain relatively uncommon and may raise genuine diagnostic and therapeutic challenges. We report the case of a 46-year-old patient who presented with a mandibular swelling evolving over six months in a context of rapidly increasing volume associated with recurrent infectious episodes. Clinical examination revealed a large mandibular swelling with cortical expansion, dental arch deformity, and submental fistulization. Imaging showed a large mandibular cystic lesion with cortical expansion and perforation, suggesting an aggressive odontogenic process. An initial biopsy performed under local anesthesia was inconclusive. The patient subsequently underwent complete enucleation with curettage under general anesthesia. Definitive histopathological examination confirmed the diagnosis of odontogenic keratocyst without evidence of malignancy. The postoperative course was marked by local infection with wound dehiscence, requiring secondary surgical management. The subsequent outcome was favorable after local debridement and close follow-up. Through this case, we discuss the clinical, radiological, and therapeutic features of large odontogenic keratocysts, as well as the importance of long-term follow-up given the high risk of recurrence.

**Keywords:** Odontogenic keratocyst; mandible; maxillofacial surgery; recurrence; odontogenic lesion; surgical treatment.

**Copyright © 2026 The Author(s):** This is an open-access article distributed under the terms of the Creative Commons Attribution **4.0 International License (CC BY-NC 4.0)** which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

## INTRODUCTION

Odontogenic keratocyst represents a distinctive entity among odontogenic lesions of the jaws. Although histologically benign, its clinical behavior is often aggressive, with sometimes considerable intraosseous extension and a significant recurrence rate reported in the literature. This duality between histological benignity and clinical aggressiveness explains the long-standing debate regarding its classification over recent decades.

Odontogenic keratocyst develops from remnants of the dental lamina and predominantly affects young male patients. The mandible is the most frequent site, particularly the angle and ramus region. Lesions may remain asymptomatic for a long period and may be discovered incidentally on radiographic examination. However, advanced forms may become symptomatic and present with swelling, facial deformity, pain, secondary infection, or, more rarely, pathological fracture.

Diagnosis is based on a combination of clinical, radiological, and histopathological findings. Therapeutic management remains debated. Several treatment strategies have been proposed in the literature, ranging from simple decompression to more aggressive surgical procedures. The choice depends mainly on the size of the lesion, its extension, the risk of recurrence, and the patient's clinical condition.

We report a case of a giant mandibular odontogenic keratocyst diagnosed at an advanced stage in an adult patient, illustrating the diagnostic and therapeutic difficulties that these lesions may pose.

## CASE REPORT

The patient was a 46-year-old man with no significant medical history, who presented with a mandibular swelling evolving over approximately six months. He reported a progressively rapid increase in size associated with intermittent painful episodes. One month before consultation, the course was marked by the

development of a submental fistula with viscous seropurulent discharge, which eventually prompted specialized consultation.

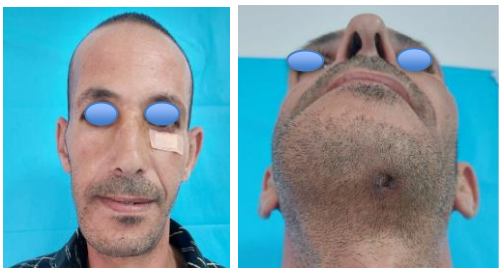

**Figure 1A-B: Preoperative clinical photographs showing the mandibular swelling in frontal view (A) and an inferior view with submental fistulization (B)**

Clinical examination revealed a large swelling involving the bilateral symphyseal and parasymphyseal mandibular region. The mass was firm, fixed to both superficial and deep planes, and appeared to be continuous with the mandibular bone. Intraoral examination showed marked cortical expansion associated with dental arch deformity and tooth loss. No initial labiomental hypoesthesia was noted.

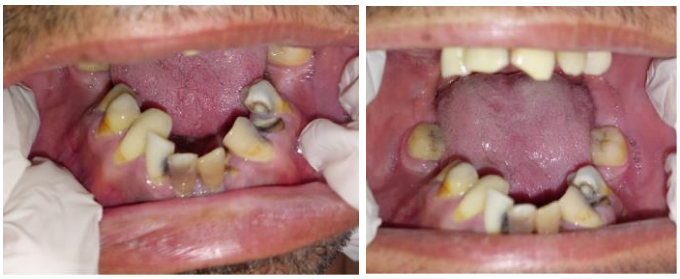

**Figure 2A-B: Intraoral views showing symphyseal and parasymphyseal cortical expansion associated with dental arch deformity**

The panoramic radiograph showed a large, extensive mandibular lesion with bony expansion and thinning of the cortical plates. Craniofacial computed tomography further defined the extent of the lesion and showed a hypodense cystic formation expanding the cortical bone, with focal areas of cortical perforation and a fistulous tract.

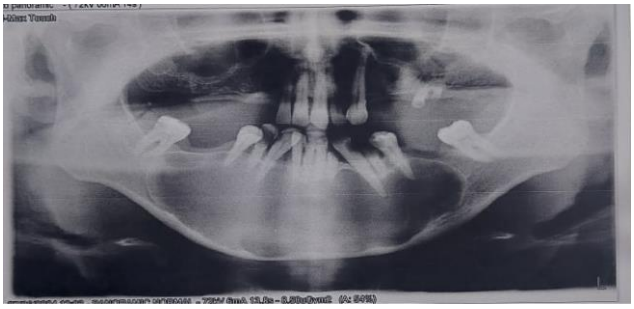

**Figure 3: Preoperative panoramic radiograph showing an expansile mandibular lesion with cortical thinning and dental arch deformity**

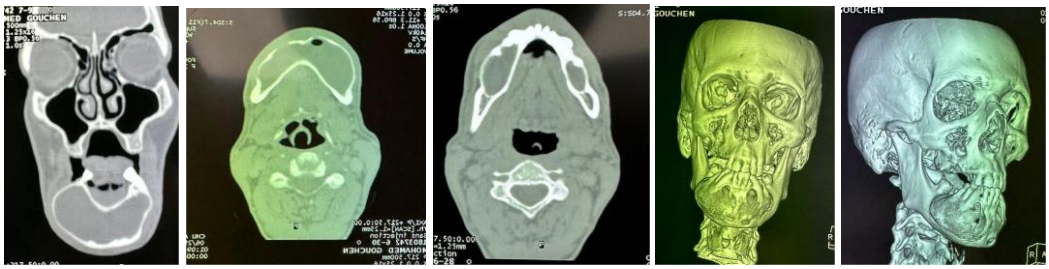

**Figure 4A-E: Craniofacial CT scan showing a large hypodense cystic mandibular lesion of the dentate portion, responsible for cortical expansion, cortical thinning, and focal cortical perforation**

Based on these clinical and radiological findings, several diagnostic hypotheses were considered, particularly ameloblastoma and giant odontogenic keratocyst. A bone biopsy with sampling of the cystic content was performed under local anesthesia. However, the initial histopathological examination did not allow a definitive conclusion regarding the exact histological nature of the lesion.

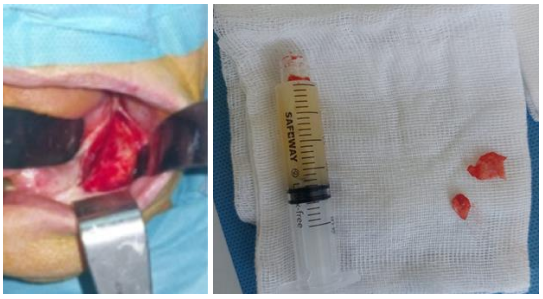


**Figure 5A-B: Biopsy performed under local anesthesia through a vestibular approach with sampling of the cyst wall and intracystic content**

Given the extensive and aggressive nature of the process, surgical management was decided. The patient underwent complete enucleation associated with curettage under general anesthesia. A vestibular approach was used, with fenestration of the external cortex, progressive dissection of the cyst wall, and complete excision of the lesion. Fistulectomy and oral dental sanitation were performed during the same surgical procedure.

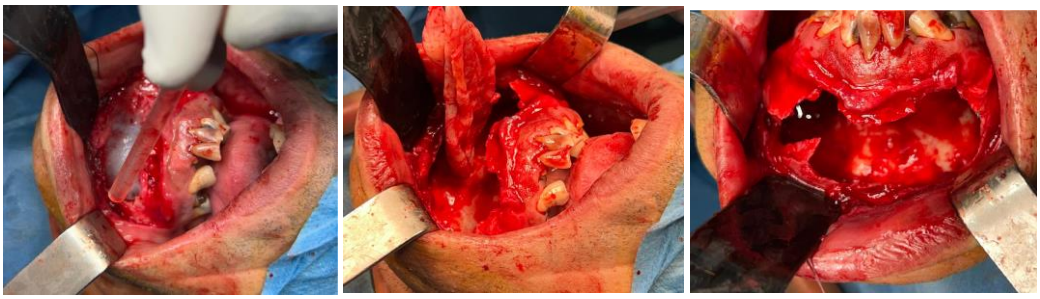

**Figure 6A-C: Peroperative views of the surgical management showing cortical fenestration (A), cyst enucleation (B), and thorough curettage of the bony cavity (C)**

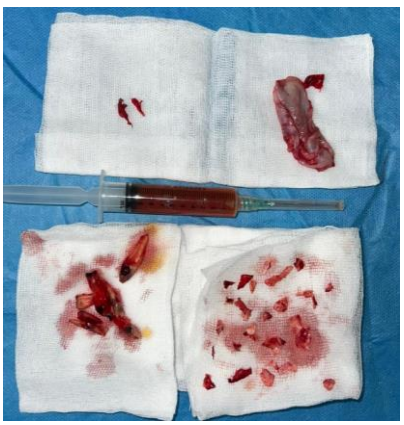

**Figure 7: Macroscopic appearance of the surgical specimen associated with cortical bone trimming and oral dental sanitation**

Definitive histopathological examination concluded to a mandibular odontogenic keratocyst without evidence of malignancy.

The immediate postoperative course was marked by wound dehiscence with local superinfection and persistence of bone debris. A second surgical procedure with irrigation, complementary curettage, and wound resuturing was required, in association with antibiotic therapy and temporary placement of a nasogastric tube to promote healing.

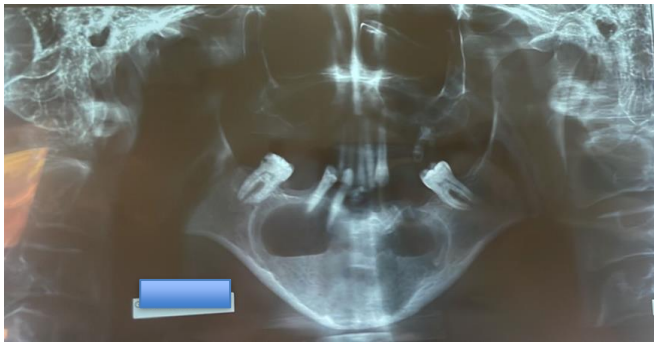

**Figure 8: Follow-up panoramic radiograph showing residual intraosseous images suggestive of persistent bone debris**

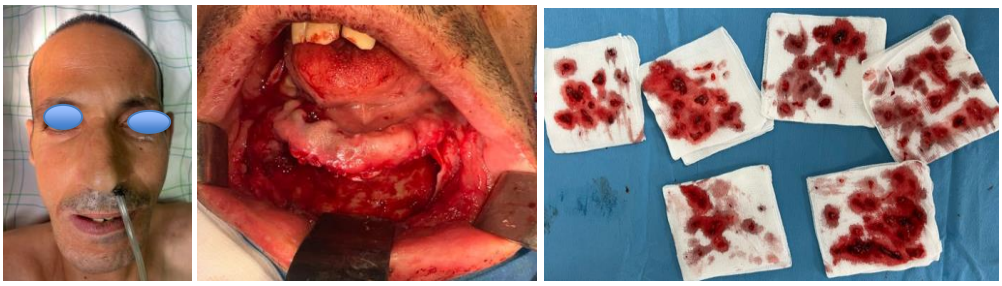

**Figure 9A-D: Per operative views during secondary surgical curettage.**

The subsequent outcome was favorable, with disappearance of pain, progressive healing of the surgical approach, and no radiological evidence of recurrence at the latest follow-up. The patient nevertheless presented with mild labiomental hypoesthesia, which required close clinical monitoring and was marked by recovery of sensitivity, as well as prosthetic rehabilitation improving masticatory function, speech, and facial morphology.

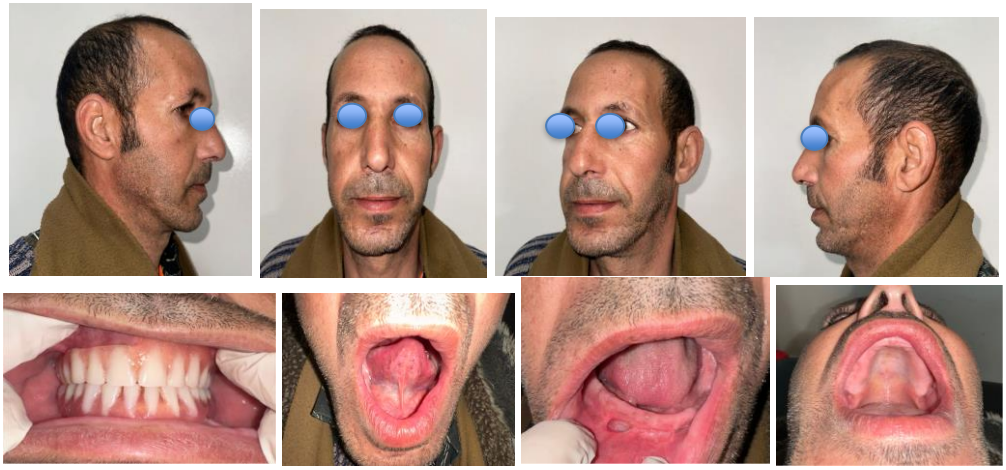

**Figure 10A-H: Clinical photographs at one-year postoperative follow-up including frontal and profile views, as well as intraoral views (mouth opening, occlusion with dental prosthesis, and tongue protrusion)**

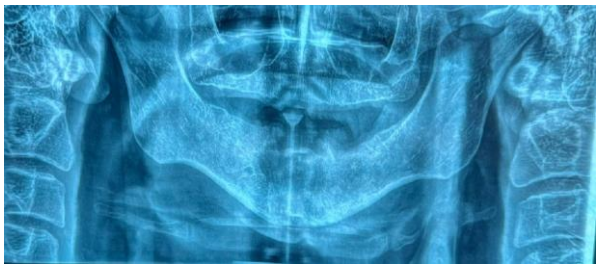

**Figure 11: One-year post operative panoramic dental radiograph showing satisfactory bone consolidation without any image suggestive of local recurrence.**

## Discussion

Odontogenic keratocyst remains a distinctive lesion among odontogenic jaw pathologies. Although it is traditionally considered benign, its clinical behavior may be particularly aggressive, with extensive bone involvement and significant functional impact. This local aggressiveness is mainly related to epithelial proliferation, the possible presence of satellite microcysts, and the fragility of the cystic capsule.

The mandible, particularly the posterior region and ramus, is the most commonly reported site in the literature. However, giant forms with extensive symphyseal involvement remain relatively rare. In our case, the large mandibular extension and cutaneous fistulization reflected an advanced stage and significant bone destruction.

Clinically, the long asymptomatic course of odontogenic keratocyst often explains delayed diagnosis. Several series report that lesions are frequently discovered incidentally during radiological assessment performed for another reason. Clinical manifestations usually appear when the lesion becomes large or is complicated by secondary infection. In our patient, pain, rapid enlargement, and fistulization were the main reasons for consultation.

Imaging plays a central role in the assessment of these lesions. The panoramic radiograph is often the first-line examination. It can reveal a well-defined radiolucent image, either unilocular or multilocular. Computed tomography remains essential in extensive forms in order to accurately assess bone extension, the condition of the cortical plates, and regional anatomical relationships. In our case, CT scan demonstrated areas of cortical perforation and a fistulous tract, supporting the aggressive nature of the lesion.

The differential diagnosis mainly includes other aggressive odontogenic lesions, especially ameloblastoma. In some cases, the clinical and radiological appearance may be very similar, making preoperative diagnosis difficult. Preoperative biopsy remains useful but may sometimes be noncontributory, as observed in our patient, particularly in the presence of significant inflammation or insufficient sampling.

Treatment of odontogenic keratocyst remains controversial. Some teams favor conservative procedures such as decompression or marsupialization, especially in young patients or in large lesions. Other authors recommend more aggressive approaches in order to reduce the risk of recurrence. Enucleation associated with peripheral curettage remains one of the most widely used techniques in accessible lesions.

In our case, the choice of enucleation with curettage was motivated by the extensive nature of the lesion, while seeking to preserve mandibular continuity as much as possible and to limit the morbidity of a more radical procedure. The postoperative infectious complication observed in our case was probably related to the size of the residual cavity and the pre-existing inflammatory context.

The main therapeutic challenge of odontogenic keratocyst remains the high risk of recurrence. Depending on the series, recurrence rates vary from 5% to 60%. This variability is mainly related to the surgical technique used and the duration of follow-up. Recurrences may occur several years after initial treatment, justifying prolonged clinical and radiological surveillance.

Through this case, we emphasize the importance of early diagnosis and appropriate management in order to limit the functional, infectious, and aesthetic complications associated with advanced forms of odontogenic keratocyst.

## Conclusion

Mandibular odontogenic keratocyst is a benign lesion whose clinical behavior may be particularly aggressive. Giant forms remain rare but may be responsible for significant bone destruction and infectious complications. Diagnosis relies on the correlation of clinical, radiological, and histopathological findings. Management must be adapted to the extent of the lesion and to the risk of recurrence. Finally, prolonged clinical and radiological follow-up remains essential given the recurrent potential of this pathology.

## References

1. Philipsen HP. Om keratocyster (kolesteatomer) i kaeberne. Tandlaegebladet. 1956; 60:963-980.
2. Barnes L, Eveson JW, Reichart P, Sidransky D. World Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours. IARC Press; 2005.
3. Johnson NR, Batstone MD, Savage NW. Management and recurrence of keratocystic odontogenic tumor: a systematic review. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;116(4): e271-e276.
4. Stoelinga PJW. The odontogenic keratocyst revisited. Int J Oral Maxillofac Surg. 2022;51(6):799-806.
5. Chen P *et al.,* Clinicopathological features and treatment outcomes of odontogenic keratocysts. Transl Cancer Res. 2022;11(8):2854-2862.
6. Fidele NB *et al.,* Recurrence of odontogenic keratocysts and prognostic factors. Med Oral Patol Oral Cir Bucal. 2019;24: e491-e501.
7. Moellmann HL *et al.,* Comparison of five treatment approaches of odontogenic keratocyst. Head Face Med. 2023; 19:6.

8. Myoung H *et al.,* Odontogenic keratocyst: review of 256 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;91(3):328-333.
9. Bande CR *et al.,* Prevalence, treatment and recurrence of odontogenic keratocyst. J Maxillofac Oral Surg. 2010;9(2):146-149.
10. MacDonald-Jankowski DS. Keratocystic odontogenic tumour: systematic review. Dentomaxillofac Radiol. 2011;40(1):1-23.