Endoscope-Assisted Enucleation of Mandibular Odontogenic Keratocyst Tumors

Antonio Romano, Giovanni D.A. Orabona,* Vincenzo Abbate,* Fabio Maglitto,* Domenico Solari,* Giorgio Iaconetta,† and Luigi Califano**

Abstract: The keratocyst odontogenic tumor (KCOT) represents a rare and benign but locally aggressive developmental cystic lesion usually affecting the posterior aspect of the mandible bone, the treatment of which has always been raising debate, since Philipsen first described it as a distinct pathological entity in 1956.

Recent studies have proposed the use of endoscope-assisted surgical technique, due to the possibility given by the endoscope of improving the effectiveness of the treatment of these lesions thanks to a better visualization of operative field and though a better understanding of the pathology. In this article, we would like to present our experience with the endoscope-assisted treatment of KCOT of the posterior region of the mandible.

From April 2000 to April 2012, 32 patients treated for KCOT were enrolled in our retrospective study: patients were divided in 2 groups according to the type of treatment, that is, 18 were treated with traditional enucleation surgery (TES), and 14 patients underwent endoscopic assisted enucleation surgery (EES).

Fischer exact test and Kaplan–Meier curves were used to compare the outcomes between the 2 focusing on the recurrence and complication rates. In the TES group, patients we found a higher recurrence rate (39%) and higher postoperative complication rate at 5-year follow-up.

Our data suggested, though, that EES seems to be a feasible alternative for the treatment of posterior mandibular KCOT. Further studies and larger series are needed to confirm these results.

Key Words: Cystic jaw lesions, endoscope-assisted enucleation, endoscope-assisted surgery, KCOT, odontogenic keratocyst

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Keratocysts odontogenic tumors (KCOTs), first described by Philipsen,[1] are benign osseous-destructive lesions that exhibit keratinization of their epithelial lining, arising in the maxilla and/or in the mandible. They arise from remnants of the dental lamina retained within the alveolar bone following tooth development. KCOTs were the third most common jaw cyst and constituted 10.2% of all odontogenic cysts.[2]

Usually, these benign lesions are asymptomatic, although they can increase their dimension due to inflammatory reaction. Sometimes, lesion diagnosis is incidental, eventually related to the occurrence of a superinfection causing pain and purulent secretion in the oral cavity. The incidence is in the early fourth decade of life, around 30 years of age.[3]

These lesions may occur in the posterior mandible in a dentigerous configuration associated with the crown of an unerupted tooth. They were often found occasionally at routine orthopanoramic radiography (OPT RX).

The treatment of these lesions is still more controversial. Indeed, the diagnosis and the management of benign lesions of the jaws is one of the most exciting topics in head and neck pathology.[1]

Several authors suggest a conservative treatment instead of an aggressive one. Conservative treatment includes enucleation of the cyst, curettage of the cavity, marsupialization, and decompression.[4] On the contrary, aggressive therapies consist of en-bloc bone resection.[5] Management of these lesions is still now controversial due to the high percentage of recurrence after surgical treatment, as reported in the pertinent literature.[6,7]

Complete removal of the cyst with its epithelium represents the therapeutic goal and could grant a lower recurrence rate. This target may be difficult to achieve by direct visualization for mandibular posterior localization.

Recent studies have proposed the use of endoscope-assisted surgery due to the possibility given by the endoscope of improving the effectiveness of the treatment of these lesions thanks to a better visualization of operative field and through a better understanding of the pathology.[8–11]

In this retrospective study, we would like to present our experience with endoscopic assisted treatment of KCOT occurring in the posterior region of the mandible. To our knowledge, this is the largest series of KCOT removed with endoscope-assisted technique, reported in the literature.

METHODS

A retrospective chart review was performed in 221consecutive patients admitted to our department presenting mandibular cystic lesion from April 2000 to April 2012. Inclusion criteria were the following:

1) Histological diagnosis of KCOT;
2) Minimum follow-up period of 36 months;
3) Lesion extending posteriorly to an imaginary line perpendicular to the tangent at the most inferior point of gonion, calculated on OPT RX (Fig. 1).

Exclusion criteria were:

1) Non KCOT osteolytic areas;
2) Relapsing lesion;
3) KCOT involving portion of the mandible anterior to our line.

A total of 32 patients met the above criteria and were enrolled in the study. Patients were divided into 2 groups according to the type

From the *Department of Maxillofacial Surgery, University of Naples "Federico II," Naples; and †Department of Neurosurgery, University of Salerno, Salerno, Italy.

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Address correspondence and reprint requests to Vincenzo Abbate, MD, Department of Maxillofacial Surgery, University of Naples "Federico II," Via Pansini 5, Naples 8013, Italy;
E-mail: abbate.maxfacc@gmail.com

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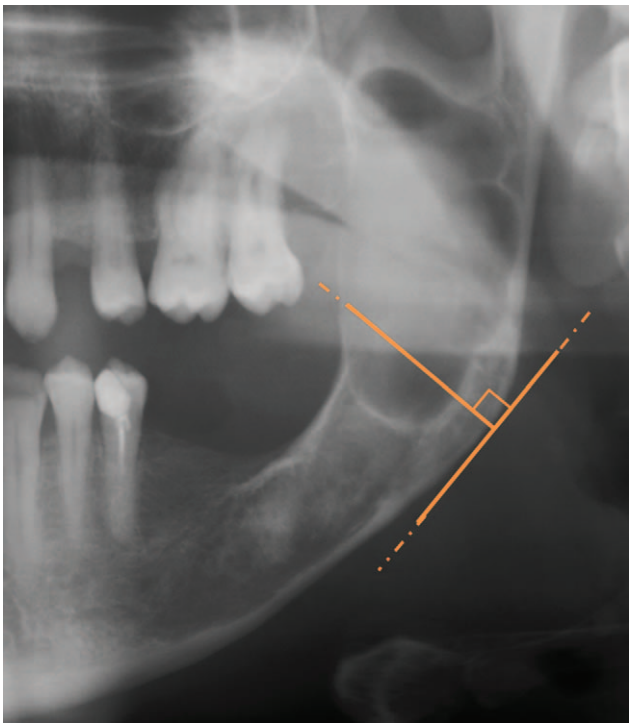

FIGURE 1. Orthopanoramic radiography showing keratocysts odontogenic tumor crossing an imaginary line perpendicular to the tangent straight at gonion.

of treatment: 18 were treated with traditional enucleation surgery (TES) and 14 patients underwent endoscopic assisted enucleation surgery (EES).

TES group patients were all treated before February 2008. From this date over, patients were treated using the endoscopic technique (EES) that was added to our surgical armamentarium.

Cysts were diagnosed at a routine OPT RX and computer tomography (CT) scan. A preoperative biopsy was performed in all the patients to confirm the KCOT nature of the lesion. Root treatment of the tooth involved in the cystic cavity has been performed prior than surgery in all the patients who did not require tooth extraction.

Endoscopic Assisted Enucleation Surgery Technique

Surgery was performed under general anesthesia. Local anesthesia with mepivacaine and adrenaline was injected intraorally at the retromolar area. Depending on the lesion size, a mucoperiostal incision was performed from the molar region to the anterior ascending ramus. Blunt tissue dissection was then accomplished subperiosteally on the lateral aspect of the mandibular ramus. If the third molar was present, we first performed the odontectomy and tooth sectioning to remove it. A bony window through cortical bone was performed to expose the lesion, whether bony erosion was not observed.

Thereafter, a 0-degree Hopkins endoscope (Karl Storz, Tuttlingen, Germany) was inserted intraorally, so that cyst removal was carried out under direct visualization. The surgeon held the endoscope with the nondominant hand while the other hand performed surgical maneuvers. Through the bony window, a 30- to 45-degree, 4-mm Hopkins scopes (Karl Storz) were introduced into the cystic cavity to achieve a panoramic 360-degree inspection of the surgical site, to ensure a complete epithelial removal (Fig. 2A).

Under endoscopic control, it was possible to identify the alveolar nerve and separate it from the cyst lining (Fig. 2B). A curved round burr was used to achieve a complete curettage of the bony cystic wall (Fig. 2C); hence, the empty cavity was irrigated with saline solution (Fig. 2D), and the wound was closed with a polyglactin absorbable 3.0 suture.

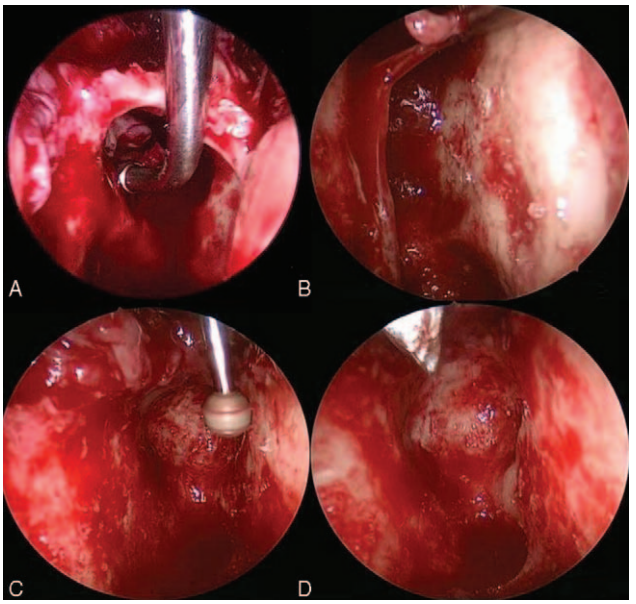

FIGURE 2. A 30-degree endoscopic view of the cavity: (A) a curved suction tube is used to detach the cyst lining from the bony walls; (B) Blakeslee Weil forceps were used to remove tissue fragments and to scrape the residual cavity; (C) a curved round burr was used to perform a complete curettage of the bony cystic wall; (D) empty cavity after surgical curettage.

Traditional Enucleation Surgery

This surgical technique consists in the removal of the entire cystic lining together with any associated overlying mucosa followed by extensive cavity curettage. To eliminate possible epithelial residues from the cyst wall that may have been left behind, gauze soaked with saline solution was used to scrap the surgical cavity. The wound was then closed in a standard fashion.

Patient's Outcome

The patient was given specific instructions for a soft diet and prescription for appropriate antibiotic and analgesic therapy. The patient had follow-up appointment every 6 months after surgery with RX OPT. A CT scan was taken once a year to assess the progressive bone healing and to evaluate the eventual residual lesion.

The disease recurrence rate for each group was assessed by the Kaplan–Meier curve. Fisher exact test was used to analyze the outcome parameters' correlation between the 2 groups.

RESULTS

A total of 32 KCOTs were admitted in our studies. Our series is constituted of 23 male and 19 female patients. The mean age was 37 years (range 16–76). Patients were followed for a minimum of 36 months postoperatively. The TES group patients had the longest follow-up period, ranging from 36 to 84 months, mean 60 months. In the TES group patients, we found a higher rate of postoperative complications: 3 patients reported accidental mandibular fracture during the first 6 months after the surgical procedure, whereas none was reported in the EES group. Moreover, in the TES group, we observed 7 patients with labial anesthesia related to mandibular

TABLE 1. Postoperative Outcome Evaluation

Postoperative Outcome	TES Group	EES Group	*P*
Mandibular fracture	3 (16.6%)	0	0.24
Mandibular nerve weakness			
Temporary	5 (27.7%)	1 (7.1%)	0.053
Permanent	2 (11.1%)	0	
Total patients	18	14	

EES, endoscopic assisted enucleation surgery; TES, traditional enucleation surgery.

TABLE 2. Patient Recurrence Rate

Follow-Up, y	TES Group (%)	EES Group
3	5 (28)	0
5	7 (39)	0

EES, endoscopic assisted enucleation surgery; TES, traditional enucleation surgery.

nerve injury. In 5 patients, there was a complete recovery 6 months postoperatively. In the EES group, on the other side, we found just 1 patient with transient mandibular nerve palsy (Table 1). The 2 groups were also compared regarding the KCOT recurrence rate. At 3 years postoperatively, a recurrence rate of 28% was observed in the TES group. This percentage increases to 39% at 5-years follow-up. No recurrence was observed in the EES group at 3 and 5 years postoperatively (Table 2). The Kaplan–Meier curve was used to report each recurrence event during the follow-up. We found a higher recurrence rate between the 22nd to the 32nd postoperative month (Fig. 3).

DISCUSSION

The KCOT treatment has been always raising debate, since Philipsen[1] first described it as a distinct pathological entity in 1956. The World Health Organization's recent classification of head and neck tumors redefined the keratocyst as a benign neoplasm, recommending the term "keratocystic odontogenic tumor" (KCOT) so that cystic jaw lesions that are lined by the orthokeratinising epithelium are now not included in the spectrum of KCOT.[12]

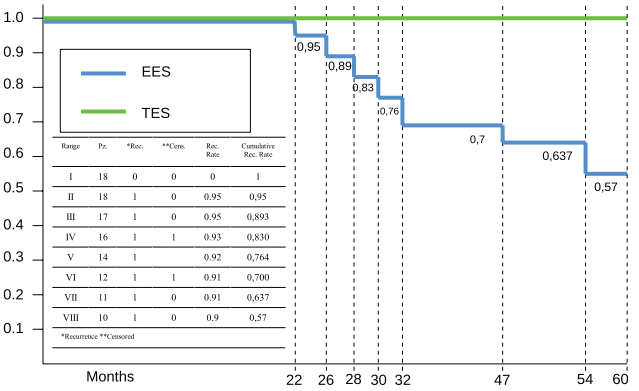

FIGURE 3. Kaplan–Meier curve demonstrating a longer recurrence-free survival period in the endoscopic assisted enucleation surgery group.

KCOT is a benign neoplasm of odontogenic origin with potential aggressive and infiltrative behavior. Considerable debate still exists regarding the recurrence rate and morbidity associated with the surgical management of this tumor. Nevertheless, the large variability and the lack of homogeneity between patients in the different studies did not allow the development of universally recognized guidelines for KCOT treatment. Several treatment options have been described in the literature, ranging from simple enucleation with or without the use of adjunctive treatment to more aggressive surgical removal. Kaczmarzyk et al,[7] in their cornerstone systematic review, analyzed a total of 1568 articles related to the treatment and the recurrence rate of KCOT. Of these, only 2 articles have met standard scientific requirements and entered the final review.[6,13] The authors described all the treatment modalities for KCOT and related recurrence rates: they found a recurrence rate ranging from 15.19% to 31.11% regardless of the treatment method. Specifically, they found that the most common treatment modality in literature was enucleation alone with a recurrence rate of 26.09%. The adjuvant intraoperative therapy with any kind of agent that destroys epithelial remnants (Carnoy solution) did not reduce significantly the recurrence percentage. They concluded that in the current literature, the treatment of choice for KCOT is still debatable. Our data disclose that that the possibility to explore the entire volume of the cavity to remove hidden epithelium fragments could tremendously reduce the risk of recurrence. Indeed, the endoscope provides a direct view of the cavity, thus allowing the visualization and removal of microscopic residual. Troulis and Kaban[14] first described the use of endoscopy to access the mandibular ramus during minimally invasive orthognathic surgery. In 2003, Suarez-Cunqueiro et al[15] reported the use of endoscopic technique to remove an ectopic third molar at the mandibular condyle area with enucleation of dentigerous cyst. Also, Kretzschmar et al[8] described the use of endoscopy for the treatment of a mandibular condyle lesion. Saia et al[11] reported the endoscopic approach for enucleation and curettage of mandibular cyst.

To date, only 1 patient with KOCT treated with endoscope-assisted technique was reported in the literature. Sembronio et al[9] described a case in which endoscopy was used to assist the enucleation and curettage of a voluminous KCOT of the mandible.

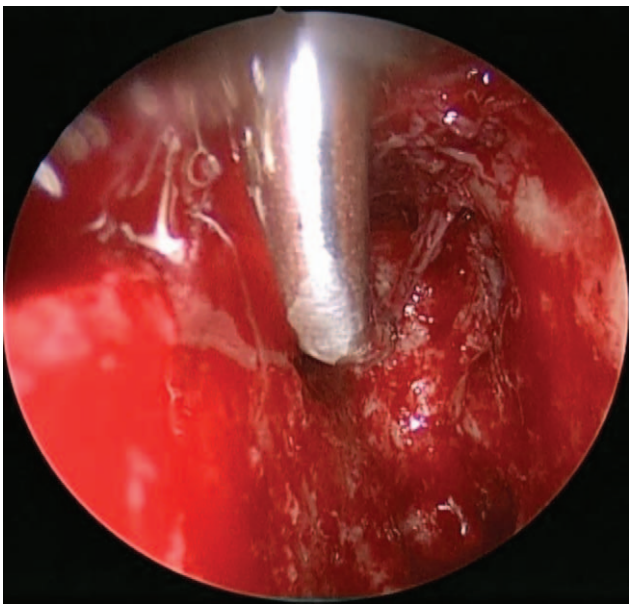

FIGURE 4. A 45-degree endoscopic view: epithelial remnants on the roof of cavity after total enucleation.

At 3-years follow-up, no clinical or radiological evidence of recurrence was noted.

In our series (EES group), we found in 10 cases (71%) the presence of epithelial remnants in the residual cavity at the endoscopic exploration after gross-total enucleation (Fig. 4). We removed the tissue and the bony wall under direct orientable endoscopic vision.

We found that endoscopic control allowed separation of the cyst lining from the nerve and the bony walls in a safer way through a very small access, allowing the preservation of the bone and thus reducing the risk of a jaw fracture and damage to the facial nerve. The analysis of the 2 groups confirmed that there was a significant difference when comparing the risk of nerve injury ($P = 0.053$ at Fisher test), whereas there was not in regards to the postoperative jaw fracture risk ($P = 0.24$ at Fisher test).

In regards to the recurrence rates, we can state that in our series, the group TES presents at 3 and 5 years a percentage of recurrence, which overlaps with rates reported in the literature, whereas the EES group presented a null recurrence rate either at 3- and 5-years follow-up. However, on one side, we should consider that most part of recurrences could occur within the first 5 to 7 years,[16–18] on the other, recurrences have been reported at 9 or even at a later stage after the initial treatment.[19] Our results seem encouraging, and endoscopic assistance in the treatment of KCOT could be considered a valid therapeutic option; however, we retain that this technique should be validated through longer follow-up and larger series.

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