Peripheral ostectomy with the use of Carnoy’s solution as a rational surgical approach to odontogenic keratocyst: A case report with a 5-year follow-up

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Abstract

Introduction. Odontogenic keratocyst (OKC) is a rare developmental, epithelial and benign cyst of the jaws of odontogenic origin with high recurrence rates. The third molar region, especially the angle of the mandible and the ascending ramus are involved far more frequently than the maxilla. The choice of treatment approach was based on the size of the cyst, recurrence status, and radiographic evidence of cortical perforation. Different surgical treatment options like marsupialization, decompression, enucleation, enucleation with Carnoy’s solution, peripheral ostectomy with or without Carnoy’s solution, and jaw resection have been discussed in the literature with variable rates of recurrence.

Case report. We presented a 52-year-old male with orthokeratinized odontogenic keratocyst. Elliptical unilocular radiolucency located in the third molar region and the ascending ramus of the mandible, 40 × 25 mm in diameter with radiographic evidence of cortical perforation at the anterior ramus border of the mandible 20 mm in diameter, was registered on orthopantomographic radiography. Surgical treatment included enucleation of the cyst and peripheral ostectomy with the use of Carnoy’s solution and excision of the overlying attached mucosa. Postoperatively, no paresthesia in the inervation area of the inferior alveolaris nerve was registered. Recurrences were not registered within 5 years post-intervention.

Conclusion. Treatment of odontogenic keratocyst with enucleation and peripheral ostectomy with the use of Carnoy’s solution and excision of the overlying attached mucosa had a very low rate of recurrence. Radical and more aggressive surgical treatments as jaw resection should be reserved for multiple recurrent cysts and when OKC is associated with nevoid basal cell carcinoma syndrome (NBCCS). Following the treatment protocol in the management of OKC and systematic and long-term postsurgical follow-up are considered key elements for successful results.

Key words: odontogenic cysts; mandible; oral surgical procedures; recurrence.

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Apstrakt

Uvod. Odontogena keratocista (OKC) je retka razvojna, epitelna, benigna cista viličnih kostiju, odontogenog porekla sa visokom stopom recidiva. Predeo donjeg trećeg molara, naročito ugla i ramusa donje vilice, zahvaćeni su znatno češće nego gornja vilica. Izbor lečenja zasnovan je na veličini cisticnih promena, postoji recidiva i postojanju kortikalne perforacije evidentirane radiografski. Različiti hirurški tretmani, kao marsupijalizacija, dekompresija, enukleacija, enukleacija sa Karnojevim rastvorom, periferna osteotomija sa ili bez Karnojevog rastvora i resekacija vilice razmatrani su u literaturi sa varijabilnim podacima o stopi pojave recidiva.

čenje odontogenih keratocista enukleacijom i perifernom osteotomijom uz upotrebu Karnojevog rastvora i eksciziju okolno zahvaćene mukoze pokazuje vrlo nisku stopu pojave recidiva. Najradikalnije i najagresivnije hirurško lečenje, kao resekcija vilice, moglo bi biti rezervisano za slučajeve čestih recidiva cista i kada je OKC udružena sa Gorlin-Golcovim sindromom. Pridržavanje terapijskog protokola u lečenju OKC i sistematično i dugotrajno postoperativno praćenje ključni su elementi uspešnog rezultata.

Ključne reči: odontogene ciste; mandibula; hirurgija, oralna; recidiv.

Introduction

Odontogenic keratocyst (OKC) was defined as a developmental, epithelial and benign cyst of the jaws of odontogenic origin. In the World Health Organization (WHO) classification of head and neck tumors from 2005, odontogenic keratocyst was reclassified and renamed to keratocystic odontogenic tumor (KCOT). Thus, this tumor was classified as a benign cystic neoplasm of the jaws of odontogenic origin. The histologic features are characterized by the presence of a thin bandlike parakeratinized or (KCOT) orthokeratinized (OKC) stratified squamous epithelium. Although KCOT with parakeratinized epithelium have aggressive behavior with potential for rapid growth, tendency for local intraosseal destruction and penetration to adjacent soft tissue, orthokeratinized odontogenic keratocyst has different characteristics and does not show aggressive behavior.

The third molar region, especially the angle of the mandible and the ascending ramus are involved far more frequently than the maxilla.

Surgically, due to extremely vulnerable epithelium, with limited surgical access, especially in the posterior part of the mandible and ascending ramus, the existence of cortical perforation, and the desire of the surgeon to protect and preserve the integrity of vital anatomical structures, it is very difficult to remove. Because of that, both keratocystic lesions have a high rate of recurrence, which contributes to special clinical approach.

Treatment of these lesions, even today, has a number of dilemmas and controversial opinions about the choice and the degree of radicalism in surgical procedure that is necessary to be applied, with the aim of eliminating the potential for recurrence, and minimizing the surgical morbidity. In other words, the current dilemmas and controversies are leading to the crucial question: When and whether aggressive and radical therapy is necessary in the treatments of this cyst/tumor?

The aim of this report was to present a patient with large orthokeratinized odontogenic keratocystic removed by enucleation and peripheral ostectomy with additional use of Carnoy’s solution and excision of the affected overlying mucosa, which was followed-up postoperatively for 5 years.

Case report

A 52 year-old male patient was admitted to the Department of Oral Surgery, Military Medical Academy (MMA) because of pain after tooth extraction which was performed elsewhere a few days ago. On the orthopanoramic radiography, an elliptical unilocular radiolucency, located in the left third molar region and the ascending ramus of the mandible, was registered. The lesion was 40 × 25 mm in diameter with radiographic evidence of cortical perforation at the anterior border of the mandibular ramus, 20 mm in diameter (Figure 1). The patient had no other clinical symptoms except pain. After the analysis of the radiographs, the patient was advised for surgical treatment after biopsy, because it is the adopted protocol for cases of the suspected KCOT. The patient accepted the proposed treatment plan.

Preoperative biopsy was performed under the inferior alveolar nerve block. The histopathological finding indicated the presence of an orthokeratinized odontogenic keratocyst (OKC). Most of the epithelium was orthokeratotic, with a mass of laminar keratin material in the lumen. The lower part of squamous epithelium was hyperplastic, due to focal inflammation.

Upon receipt of the histopathological findings, the patient was advised for hospitalization and surgery to remove...

the existing lesion under general anesthesia. In the Clinic for Maxillofacial, Oral Surgery and Implantology, MMA, all the necessary laboratory analysis for surgery under general anesthesia were made, and at the Institute of Pharmacy, MMA, Carnoy solution was prepared according to the following prescription: absolute alcohol (6 mL), chloroform (3 mL), glacial acetic acid (1 mL) and ferric chloride (1 g).

Surgical procedure was started with incision along the anterior border of the left mandibular ramus, then over the alveolar crest to the gingival margin till the tooth 35, with vertical relaxing incision down to the fornx in this region. After uplifting the mucoperiostal flap and separation of the masseter muscle and the pterygoid medial muscle attachments defect of the anterior border of ramus about 20 mm in diametar was seen (Figure 3). The defect was slightly wid-

ened caudally at the lateral aspect of the mandibular ramus, in order to approach the lesion. It was completely enucleated, a defect rinsed with saline, and a sterile gauze swab pre-
soaked with Carnoy’s solution was placed in the lumen of the defect and left there for 3 minutes. Then, the lumen of the defect was re-rinsed with saline to enable sight of the cystic wall remains, which were dark brown colored and fixated, enabling their complete removal. After that, a peripheral os-
tectomy in the caudal and cranial direction was performed and the overlying attached mucosa was excised. After repeated rinsing with saline, the defect was buffered with the iodine-vaseline gauze and the wound was cuturated.

Postoperatively, the patient was prescribed intrave-
nous antibiotics: ceftriaxon 2 g once daily and metroni-
dazol 0.5 g three times daily, for the following seven days. The patient was regularly controlled, every day. The whole iodine gauze packing was removed at the third postoperative day and the sutures were removed after 7 days. No par-
esthesia in the inervation area of the inferior alveolaris nerve was present, as well as any other postoperative comp-
lications. The patient was followed up regularly, and after a 5-year period, orthopantomographic radiography was done again (Figures 4 and 5).

Discussion

OKCs are primary jaw bone lesions, but it is interesting to note that these cysts can occur in highly atypical places. Ery-
ilmaz et al. showed the occurrence of OKC in the temporo-
mandibular joint, while Yih and Krump registered such cyst in nasopalatinal duct as a very rare occurrence. Also, some authors reported a case of peripheral OKC, outside the jaws, in the gingival tissue or even in the buccal mucosa.

OKC treatment involves surgical approach only. Gener-
ally, treatments are classified as conservative or radical (ag-
gressive). Conservative methods include: simple enucleation (with or without curettage), marsupialization with subse-
quent enucleation, and decompression with subsequent enu-
cleation. Aggressive methods include: cryotherapy, the use of Carnoy’s solution, peripheral ostectomy and resection of the jaw with or without immediate reconstruction of a defect with corticocancellous iliac crest bone allografts.

The most significant and important therapeutic prob-
lem, related to these cysts is the high rate of recurrence after initial surgical treatment. Boyne et al. and Chemli et al. reported the frequency of recurrences up to 60%, while some authors have registered recurrence after 25 years, or even 41 years after the first surgical intervention. The most
common reason for recurrence is incomplete removal of cyst walls or epithelial islands and/or microcysts, in the peripheral bone and the overlying soft tissue. Stoeleinan stated that in approximately 50% of islets of a cyst wall and epithelium and/or microcysts remain in the overlying soft tissue, particularly in cases of cortical perforation.

Literature data suggest that recurrences appear most frequently within the first 5 to 7 years after initial surgery. Chirapathomsakul et al. reported the frequency of recurrence by as much as 71.4% in the first five years. Also, Pitak-Arnroppet al. reported that the thirty-two of the 37 recurrences occurred within 5 years. However, as stated by Crowley et al., more than 25% of recurrences occur after 9 years or even later after the first surgery.

It seems that age of the patient, localization of OKC and site of involvement, its histological type (orthokeratosis or parakeratosis), do not significantly affect the incidence of recurrence. In other words, the recurrence rate mainly depends on the applied surgical methods. However, the choice of surgical treatment depends on several factors, such as the size of the lesion, frequency of recurrence and radiographic evidence of cortical perforation. Pitak-Arnroppet al. reported that the recurrence was found in 28 patients and 11 of these had cortical perforation at the time of first presentation. Because of that, a crucial point in the treatment of OKCs is an adequate or appropriate choice of surgical method.

However, the treatment of OKC remains controversial. There are different data on the incidence rate of recurrence after different surgical methods of treatment. There are several explanations of such diversity. First of all, OKC is extremely rare, so the number of samples used in study is usually little. Furthermore, most studies have retrospective nature, often lacking valid and adequate clinical data and control groups. Finally, a period of follow-up of operated patients is highly variable in different studies.

Although it is stated that patient age does not influence the recurrence, it can be a very important factor in the choice of surgical treatment. The choice of conservative methods, despite high rates of recurrence, may be justified if there is a risk of injury to the surrounding anatomic structures, especially in young patients, because the use of aggressive surgical techniques in children can cause disturbances in the growth and development of jaws and teeth.

However, although some authors report positive clinical experience using conservative methods (marsupialisation, decompression methods and simple enucleation) in the surgical treatment of these cysts, we believe that it cannot be the definitive surgical method to treat OKC due to extremely high rates of recurrence, which is the attitude of many authors. Also, some authors suggest that enucleation, without Carnoy’s solution, provides clinically acceptable and satisfactory results with a recurrence rate of 13.3% or 26%. Tolstunov and Treasure believe that the histological diagnosis of OKC does not indicate immediate rehearsal of surgery because only 25%–50% of the cases recur after simple enucleation, and suggest that a “wait and see” protocol should be applied. However, many authors suggest that simple enucleation as surgical option without Carnoy’s solution is not adequate because of high recurrence rates of 50% or 54.5% or to 62%.

Recurrence rate after surgical treatment of these lesions is certainly a big therapeutic problem. Therefore, radical surgical intervention with the use of Carnoy’s solution and radical removing the soft tissue is sometimes suggested. In our view, for lesions with such aggressive behavior, regardless its “benign” nature, a more aggressive and radical surgical approach is needed. However, our opinion is that resection of the jaw, as the most radical and aggressive therapeutic option, should be used in the treatment of OKCs only in cases of frequent recurrences (more than three) and in situations when OKC is associated with nevoid basal cell carcinoma syndrome. There are two major reasons for this: resection of the jaw causes significant functional and aesthetic problems to the patient and less aggressive surgical methods, such as peripheral osteotomy with the use of Carnoy’s solution, provide an extremely low rate of recurrence, without functional and aesthetic problems.

Peripheral osteotomy was defined as a peripheral bone reduction with powered hand-piece and rotary instruments, done after enucleation of the lesion. This method can be combined with the use of Carnoy’s solution, which acts as a cauterizing agent, causing denaturation of organic molecules. Its penetration into the tissue results in rapid local fixation and hemostatic action. Some authors suggest that the major disadvantages of Carnoy’s solution are its systemic toxicity and local caustic effect resulting in a damage of vital anatomical elements (especially nerves). However, Blanas et al. state that application of Carnoy’s solution to cyst cavity for 3 min after enucleation should not damage the inferior alveolar nerve.

Certainly, the use of Carnoy’s solution as an adjunct measure in surgical treatment of OKC and its correct application is of non questionable importance. This method can be combined with the use of Carnoy’s solution, which acts as a cauterizing agent, causing denaturation of organic molecules. Its penetration into the tissue results in rapid local fixation and hemostatic action. Some authors suggest that the major disadvantages of Carnoy’s solution are its systemic toxicity and local caustic effect resulting in a damage of vital anatomical elements (especially nerves). However, Blanas et al. state that application of Carnoy’s solution to cyst cavity for 3 min after enucleation should not damage the inferior alveolar nerve.

A recent evidence reveals that most of epithelial islands and/or microcysts, as the most crucial factors for recurrence, are in the overlying mucosa that contacts a cyst, especially in the mandibular retromolar area. Therefore, the essence of Carnoy’s solution use lies in its influence on epithelial islands and microcystic formations; a consequent peripheral osteotomy with excision of the overlying attached mucosa completely eliminates the presence of epithelial changes.

The results of studies that compared the incidence of recurrence of different surgical techniques in OKC showed that in cases of peripheral osteotomy combined with Carnoy’s solution no recurrences were noted in ten years and more postoperatively.

Conclusion

In the presented case operated by enucleation of the lesion with peripheral osteotomy and the use of Carnoy’s solution, including excision of the overlying attached mucosa, no
reurrence was noted in a 5 year postoperative period. Therefore, we believe that the choice of this surgical method in treating odontogenic keratocyst is a rational approach to reduce recurrence. However, due to the possibility of late recurrence, a long-term postoperative follow-up is needed to confirm a successful result of the OKC treatment.

REFERENCES


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