Odontogenic keratocyst of the mandible: A case report and literature review

Zębopochodna torbiel rogowaciejąca żuchwy – opis przypadku i przegląd piśmiennictwa

Kamil Polak[1,B–D,F], Magdalena Jędrusik-Pawłowska[1,A,C–F], Bogna Drozdzowska[2,B,C,E,F], Tadeusz Morawiec[3,A,B,E,F]

[1] Department of Maxillofacial Surgery, St. Barbara Provincial Specialist Hospital No. 5, Sosnowiec, Poland
[2] Department of Pathology, School of Medicine with the Division of Dentistry in Zabrze, Medical University of Silesia, Poland
[3] Division of Dental Surgery, Department of Craniomaxillofacial Surgery and Oral Surgery, School of Medicine with the Division of Dentistry in Zabrze, Medical University of Silesia, Bytom, Poland

A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;
D – writing the article; E – critical revision of the article; F – final approval of the article

Dental and Medical Problems, ISSN 1644-387X (print), ISSN 2300-9020 (online) *Dent Med Probl.* 2019;56(4):433–436

Address for correspondence
Kamil Polak
E-mail: kamilpolak88@gmail.com

Funding sources
None declared

Conflict of interest
None declared

Received on April 30, 2019
Reviewed on June 2, 2019
Accepted on July 12, 2019

Published online on October 28, 2019

Abstract

Based on a literature review, we analyzed the World Health Organization (WHO) classification and the treatment algorithm for the odontogenic keratocyst (OKC), formerly referred to as keratocystic odontogenic tumor (KCOT).

The KCOT reclassification from benign odontogenic tumors to odontogenic developmental cysts resulted from the emergence of new evidence regarding their morphogenesis and biological behavior. The authors of the most recent 2017 classification do not provide specific guidelines for OKC. Nevertheless, it has been observed that conservative surgical management is not necessarily associated with recurrences characteristic of neoplastic disease.

The aim of this paper was to present the effective management strategy for a local recurrence that developed following conservative OKC enucleation in a 53-year-old patient. The treatment for recurrence consisted of enucleation, marginal osteotomy and augmentation with a cancellous bone graft harvested from a tibial tuberosity. A 6-year observation period (clinical and radiological monitoring) revealed normal bone regeneration and no evidence of recurrence.

The algorithm applied in our center for the treatment of OKC/KCOT was compared with the management strategies proposed by other authors.

Key words: odontogenic keratocyst, WHO classification, autogenic bone graft, recurrence

Słowa kluczowe: zębopochodna torbiel rogowaciejąca, klasyfikacja WHO, autogenny przeszczep kości, wznowa

Cite as
Polak K, Jędrusik-Pawłowska M, Drozdzowska B, Morawiec T. Odontogenic keratocyst of the mandible: A case report and literature review. *Dent Med Probl.* 2019;56(4):433–436. doi:10.17219/dmp/110682

DOI
10.17219/dmp/110682

Introduction

The term 'odontogenic keratocyst' was first used in 1956 to describe an odontogenic cyst lined with keratinized stratified squamous epithelium. In 1992, the World Health Organization (WHO) introduced the term 'odontogenic keratocyst', synonymous with 'primordial cyst', to denote benign cysts of odontogenic origin and specific histological appearance. However, in 2005, considering a high risk of recurrence, aggressive clinical course, mutations in the tumor suppressor gene (*PTCH1*), the occurrence of satellite cysts, and the association with the Gorlin–Goltz syndrome, WHO reclassified this pathology as a benign keratocystic odontogenic tumor (KCOT).[1,2]

In 2017, though, WHO released a new classification of head and neck tumors. As there was insufficient evidence to categorize the abovementioned pathology as a neoplastic lesion, KCOT was moved back into the cyst category under the name of odontogenic keratocyst (OKC). However, the term 'keratocystic odontogenic tumor' is still in use. The authors of the 2017 classification do not specifically recommend any strict guidelines for OKC treatment. Nevertheless, it has been observed that conservative surgical management is not necessarily associated with recurrences characteristic of neoplastic disease.[1–3]

The aim of this paper was to analyze a 2-stage surgical intervention for OKC in a 53-year-old patient. We also analyzed a single-stage treatment for recurrence (which developed 2.5 years after the initial therapy), consisting of enucleation with marginal osteotomy and augmentation with a cancellous bone graft harvested from a tibial tuberosity.

Case report

A 53-year-old hypothyroid – but clinically euthyroid – patient reported to the Outpatient Department of Dental Surgery at the University Dental Centre of the Medical University of Silesia in Bytom, Poland, for the diagnosis and treatment of a 2-chamber osteolytic lesion within the mandibular body (teeth 42–36), accidentally detected on a dental X-ray.

Extraoral examination revealed left facial asymmetry, but no submandibular lymphadenopathy. The following anomalies were also found: inactive fistula within the mucous membrane of the alveolar socket (tooth 45); mandibular body bulging (teeth 31–35); and bundle bone thinning characteristic of periapical cysts. Diagnostic imaging demonstrated a 2-chamber osteolytic lesion in the area of teeth 42–36. Teeth 42, 41, 31, 32, 33, 34, and 35 remained within the cystic lumen (Fig. 1).

The preparation for surgery consisted of the endodontic treatment of teeth 42, 41, 31, 32, and 33, and the extraction of teeth unsuitable for routine treatment (34 and 35). Then, intraoral cyst enucleation was performed (the area of teeth 33–36) and the root apex of tooth 33 was resected. Subsequently, cyst decompression was carried out in the area of teeth 42–32. A cyst wall specimen was collected for histological examination. Based on the pathology reports, an initial diagnosis of mandibular body OKC was provided, and the decision was made to completely evacuate the lesion. A month later, cystectomy was performed in the area of teeth 42–32 with the apicectomy of teeth 42, 41, 31, and 32. Histological examination confirmed the initial diagnosis of OKC, a cyst with a connective tissue wall lined with parakeratinized stratified squamous epithelium (Fig. 2).[4–6]

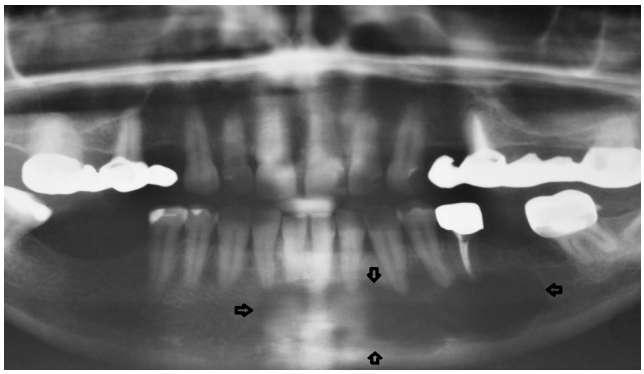

Fig. 1. Pre-treatment orthopantomogram showing a polycyclic osteolytic lesion in the area of teeth 42–36 (pointers)

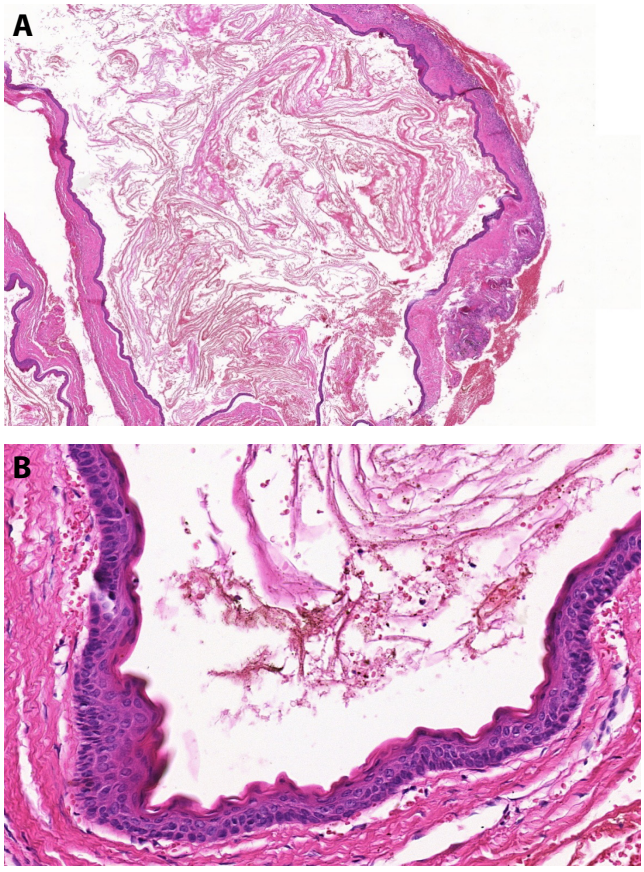

Fig. 2. A – cyst with a connective tissue wall lined with keratinized stratified squamous epithelium; the lumen contains a keratinaceous mass (hematoxylin and eosin (H & E); ×5 magnification); B – cyst with a connective tissue wall lined with parakeratinized stratified squamous epithelium; the lumen contains a keratinaceous mass (H & E; ×65 magnification)

The patient continued periodic monitoring at the Outpatient Department of Dental Surgery. A follow-up appointment 2.5 years after treatment revealed a local recurrence, an osteolytic lesion within the mandibular body between teeth 42 and 35 (Fig. 3). The patient was admitted to the Department of Maxillofacial Surgery at St. Barbara Provincial Specialist Hospital No. 5 in Sosnowiec, Poland, for radical surgery. Following the preparation for surgery, the lesion was resected along with a disease-free bone margin; re-apicectomy of teeth 42, 41, 31, 32, and 33 was also performed. The residual post-cystic cavity was augmented with a cancellous bone graft harvested from a left tibial tuberosity. Histological examination confirmed the recurrence of OKC; the bone margins were negative.

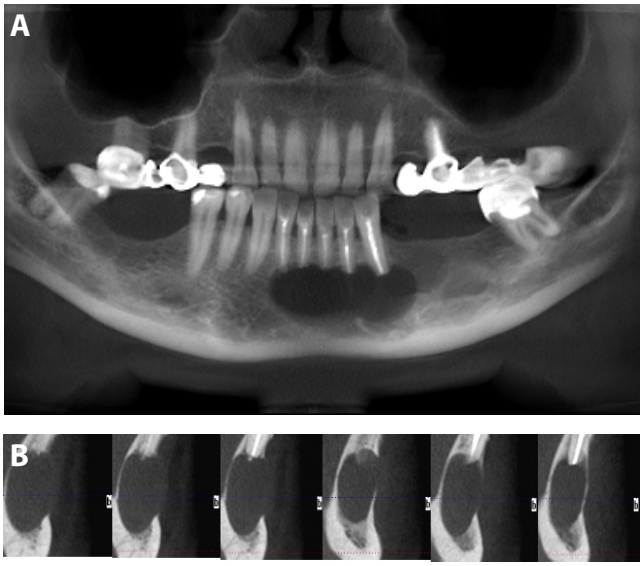

Fig. 3. A – orthopantomogram taken 2.5 years after the conservative surgical intervention, showing a mandibular keratocyst recurrence in the area of teeth 42–35; B – selected coronal sections of cone-beam computed tomography (CBCT) 2.5 years after the conservative surgical intervention, showing well-defined oval osteolysis beneath the previously resected root apices of teeth 42, 41, 31, 32, and 33 (keratocyst recurrence)

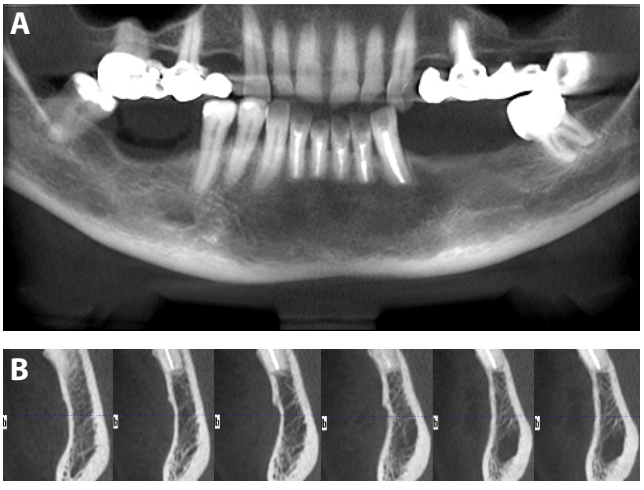

Fig. 4. A – follow-up orthopantomogram taken 4 years after marginal resection with the complete enucleation of the keratocyst in the area of teeth 42–35, showing normal bone regeneration; B – selected coronal sections of cone-beam computer tomography (CBCT) 4 years after marginal resection

A follow-up X-ray and cone-beam computed tomography (CBCT) taken 4 years after marginal resection showed normal bone regeneration and no recurrence (Fig. 4).

The resected teeth (42, 41, 31, 32, and 33) demonstrated satisfactory stability within the dental alveoli, and hence served as sufficient support for frame dentures, which the patient continues to use (Fig 5).

The patient continues with annual follow-up appointments consisting of clinical and radiological monitoring of the post-cystic bone defect. A follow-up orthopantomogram taken 6 years after radical keratocyst resection demonstrated a complete remodeling of the autologous bone transplant (Fig. 6).

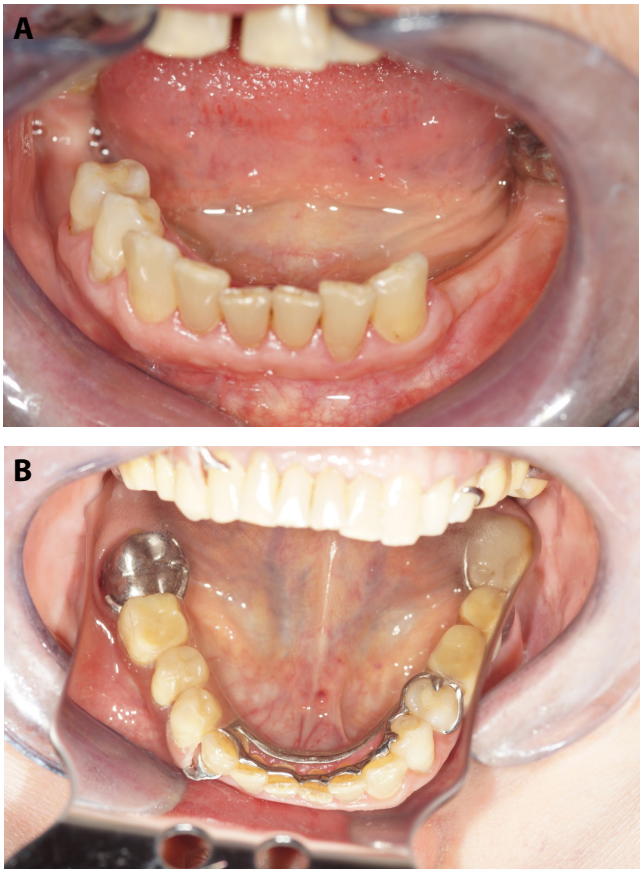

Fig. 5. Intraoral view 6 years after keratocyst resection and augmentation with a cancellous bone graft harvested from a left tibial tuberosity

A – no clinical features of recurrence; B – no evidence of mandibular body deformities; wing defects restored with frame dentures.

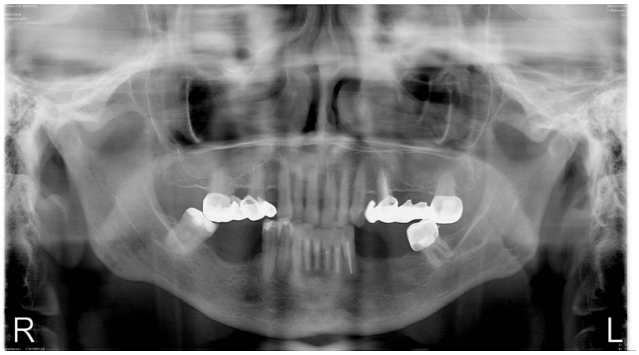

Fig. 6. Follow-up orthopantomogram taken 6 years after keratocyst resection and augmentation with a cancellous bone graft harvested from a left tibial tuberosity: Complete bone remodeling within the post-cystic area

Discussion

The reclassification of OKC from benign odontogenic tumors to odontogenic developmental cysts resulted from the emergence of new evidence regarding their morphogenesis and biological behavior. For example, it has been confirmed that a mutation of the *PTCH1* gene is not OKC-specific, as it also occurs in follicular cysts. Depending on whether the lesion is sporadic or associated with the Gorlin–Goltz syndrome, the *PTCH1* alterations are observed in 30–85% of OKCs. It has also been found that OKCs tend to regress following decompression or marsupialization, and that their lining spontaneously undergoes transition into normal oral epithelium. All of these features disqualify this clinical entity from being included in the category of neoplastic lesions and justify changing the name of the lesion to OKC.[1]

The abovementioned changes in nomenclature have had an impact on the strategy for OKC treatment, which mainly depends on the patient's age and compliance as well as the size and location of the lesion.[1] Bone resection offers the lowest recurrence rates. Segmental resection (removal of a bone segment without maintaining bone continuity) and marginal resection (removal of the lesion and a margin of uninvolved bone) are associated with recurrence rates ranging from 0% to 8.4%.[1,7–10] Despite their high efficacy, these interventions should be limited to multi-chamber lesions. Our patient presented with a recurrence 2.5 years after the initial conservative surgical intervention, hence the decision was made to perform marginal resection including a small bone fragment for histological examination.

Low recurrence rates are also characteristic of tumor enucleation with chemical curettage or cryodestruction. Chemical curettage consists in the application of Carnoy's solution (60% ethanol, 30% chloroform, 10% glacial acetic acid, and 1 g of ferric chloride) into the cyst cavity; the solution induces superficial tissue necrosis and helps eliminate the tumor remnants. In cryodestruction, the bony cavity is filled with liquid nitrogen. The recurrence rates are 14.5% and 11.5%, respectively.[1,7–10]

Conservative treatment, i.e., OKC decompression followed by enucleation or marsupialization (transformation of the cyst into an open pouch, allowing continuous drainage) is not recommended due to very high recurrence rates (17–56%).[1,7–10] Decompression should be followed by enucleation with chemical curettage, cryodestruction or marginal osteotomy, as it was in the case of our patient. The reported recurrence rate for 2-stage treatment is 14.6%.[1,7–10]

We decided on 1-stage radical surgery consisting of marginal resection and augmentation of the post-cystic defect with a cancellous bone graft harvested from a left tibial tuberosity. The treatment resulted in excellent mandibular bone regeneration. We have not come across any literature reports on similar augmentation methods following OKC enucleation. It should be noted that this treatment allowed jawbone continuity to be maintained, and the teeth and the inferior alveolar nerve to be preserved. Naturally, there is some risk of disease recurrence in allogenic and autologous bone grafts, as reported by Tolstunov and Treasure and DeGould and Goldberg.[11,12] We would not define our method as superior to other treatment options, but it is undoubtedly worth recommending. An analysis of OKC recurrence with a larger sample size is needed, which we plan to be the subject of further research.

The patient's attitude and compliance with follow-up appointments (especially during the first 6 years after surgery, when the risk of recurrence is the highest) are prerequisites for successful treatment.[7] Based on our findings, we conclude that patients diagnosed with OKCs should be regularly monitored at outpatient clinics, including follow-up orthopantomography and CBCT – at least once a year and for 6 years following the surgical intervention.

ORCID iDs

Kamil Polak https://orcid.org/0000-0001-9033-3113
Magdalena Jędrusik-Pawłowska https://orcid.org/0000-0002-8339-7618
Bogna Drozdzowska https://orcid.org/0000-0002-2287-6842
Tadeusz Morawiec https://orcid.org/0000-0002-2283-5784

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