

原文題目(出處)：	Treatment of a patient with large keratocystic odontogenic tumour in the mandible: case report with literature review. Oral Surg 2012;5:1-6.
原文作者姓名：	I. Mojsa, J. Stypulkowska, T. Kaczmarzyk, K.Okon, M. Zaleska
通訊作者學校：	Departments of Oral Surgery, Pathomorphology; Jagiellonian University Medical College, Krakow, Poland
報告者姓名(組別)：	H組 楊舜捷
報告日期：	2012/04/09

內文：

Abstract

- ◆ Presented heretofore is the case of a 26-year-old male suffering from a keratocystic odontogenic tumour that exhibits progressive development in the mandible.
- ◆ During our evaluation, the patient underwent numerous diagnostic and therapeutic procedures to restore his normal appearance and function.
- ◆ The keratocystic odontogenic tumour(KCOT)
 - formerly classified as a parakeratinised type of cystic lesion named odontogenic keratocyst (OKC)
 - reclassified in 2005 by the WHO as a neoplastic lesion.
 - Currently,the KCOT is classified as a benign unicystic or multicystic intraosseous tumour of odontogenic origin, possessing a characteristic lining consisting of parakeratinised stratified squamous epithelium
 - with the potential for aggressive, infiltrative behaviour.

Case report

- ◆ An 18-year-old male
- ◆ 1. a painful swelling developed in the submental region
- 2. a progressive displacement of lower teeth coupled with mandibular body distension



Figure 1 Preoperative intraoral picture showing lower teeth experiencing displacement and distension of the mandibular body, ranging from the area of the second left premolar to the second right molar.

- ◆ Clinical examination:
 1. painful swelling of the submental region,
 2. Enlargement of the submental lymph nodes
 3. lower teeth experiencing displacement
 4. distension of the mandibular body, ranging from the area of the second left premolar to the second right molar

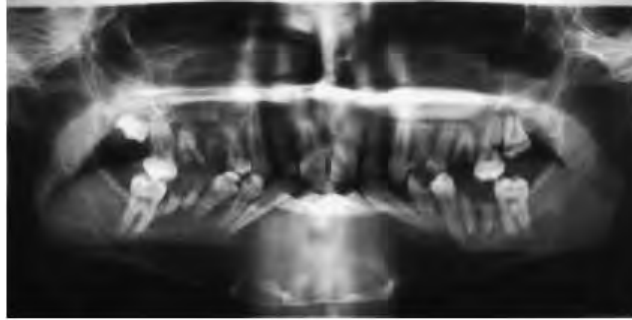


Figure 2 Preoperative orthopantomogram showing large multicystic lesion distending the mandibular body, spanned from second left premolar to second right molar.

◆ Pantomographic radiography indicated:

1. a large multicystic lesion exhibiting sharply defined borders distending the mandibular body.
2. The range → from second left premolar to second right molar
3. No root resorption, all teeth remained vital

◆ Fine-needle aspiration biopsy:

1. abundant purulent fluid
2. Bacteriological → abundant growth of oral streptococci
3. Cytological → a concentrated protein liquid with leukocytes

The surgical procedure was performed in two stages:



Figure 3 Intraoperative picture showing decompression with sample tissue biopsied for histopathological assessment.

Initial surgical stage:

→ decompression under local anaesthesia
 → with sample tissue biopsied for histopathological assessment and the affected bone cavity packed with iodoform gauze

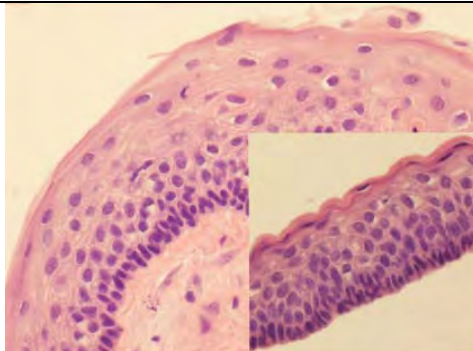




Figure 4 Histopathology. Original material: a cyst lined with seemingly non-keratinising epithelium is visible. Focal parakeratosis is present, however (inset). H + E, lens magnification 40x.


★ Histopathological assessment


→ a parakeratinised type of OKC

 <p>Figure 5 Intraoral picture showing an acrylic obturator-type appliance inserted into the bone cavity.</p>	<p>→remove gauze packs →<u>an acrylic obturator-type appliance</u> was constructed and inserted into the bone cavity.</p>
---	--

- ◆ Systematic and long-term post-first stage surgical follow-ups focused on rebuilding bone structure evaluated with imaging studies
- ◆ with the obturator, being regularly corrected as the bone rebuilding progressed.

 <p>Figure 6 Orthopantomogram (19 months after decompression) showing the lesion ranging the mandibular body from right canine to second left incisor.</p>	<ul style="list-style-type: none"> ◆ 19 months following decompression: →<u>the lesion had decreased immensely,</u> →its range spanned <u>from right canine to second left incisor</u>
--	---

 <p>Figure 7 Intraoral picture (19 months after decompression) before starting an orthodontic treatment.</p>	<ul style="list-style-type: none"> ◆ An obturator was eliminated, and <u>orthodontic treatment</u> was started so as to produce future satisfactory occlusion.
--	---

 <p>Figure 8 Intraoral picture (62 months after decompression) showing shallowness of the bone cavity in the medial region of the mandible and good results of the orthodontic treatment.</p>	<ul style="list-style-type: none"> ◆ 62 months following decompression: →shallowness of the bone cavity
---	---

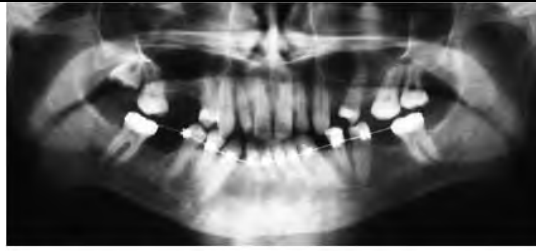


Figure 9 Orthopantomogram. Sixty-two months after decompression.

◆ The second stage of surgical:

→consisting of enucleating the residual mass of the lesion under local anaesthesia



Figure 10 Orthopantomogram (7 years after decompression and 2 years following enucleation) revealing the recurrence lesion, distending the mandibular body area spanned from right canine to second right premolar.

◆ 7 years after decompression and 2 years following enucleation:
 →an asymptomatic recurrence
 →a small unicystic lesion with sharply defined borders distending the mandibular body area
 (from right canine to second right premolar)

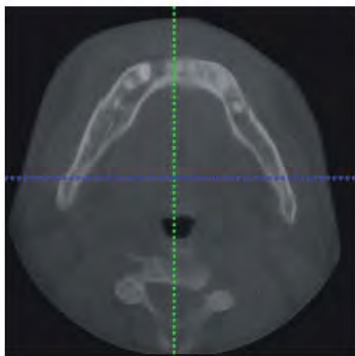


Figure 11 Preoperative computed tomography scan showing the tumour located in the proximity of the right first premolar with its lingual expansion as well as the mandibular body distention in buccal direction.

◆ Preoperative computed tomography (CT) scans:
 →the lesion located mainly in the proximity of the right first premolar with its lingual expansion
 →as well as the mandibular body distention in buccal direction

◆ The recurrence of the lesion required the patient to undergo surgical treatment:
 →the lower right first premolar was extracted to facilitate the radical removal of the lesion.

→The lesion was enucleated with peripheral bone curettage with rotary instruments.

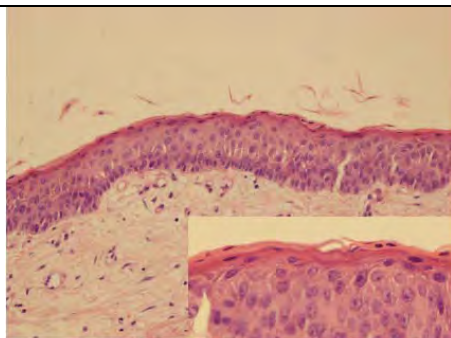


Figure 12 Histopathology. The squamous epithelium covers fibrous tissue with some blood vessels. On higher magnification, keratinising layer with dark, pyknotic nuclei (parakeratosis) is evident. H + E, lens magnification 20x, insert lens magnification 40x.

◆ Post-operative histopathological assessment confirmed the presence of a KCOT

Discussion

- ◆ KCOT:
 1. highly recurring infiltration pattern into surrounding tissues
 2. a slight predilection towards male
 3. commonly occur in the second or third decade of life
 4. typically located in the posterior region of the mandible
- ◆ Therapeutic approaches in different studies:
 - from marsupialisation and enucleation,
 - which may be combined with adjuvant therapy (Cryotherapy and Carnoy's solution)
 - to marginal or radical resection
- ◆ Decompression, followed by delayed enucleation of the residual mass of the tumour:
 - hinders a complete removal of the entire epithelial lining during the first stage of treatment.
 - may consequently lead to a continuous proliferation of the epithelium, potentially facilitating recurrence or malignisation.

Piattelli et al.	<ul style="list-style-type: none"> ◆ the <u>immunohistochemical</u> expression of the <u>B-cell lymphoma 2 bcl-2 protein</u> prove that the immunoprofile of KCOT varies from odontogenic cysts. ◆ The results of the conducted study indicate that KCOT, juxtaposed with odontogenic cysts →demonstrate bcl-2 positivity in the basal cell layer. ◆ A literature review suggests that →<u>bcl-2 is rarely</u> expressed by some basal keratinocytes of <u>normal epithelium</u>. ◆ Therefore, <u>bcl-2 protein could be useful to differentiate KCOT lining from normal epithelium</u>.
Pogrel and Jordan	<ul style="list-style-type: none"> ◆ bcl-2 negativity in normal oral mucosa and in the KCOT lining, present after decompression
August et al.	<ul style="list-style-type: none"> ◆ the epithelium of KCOTs after decompression and reported differentiation of the epithelium →with 64% of the patient presenting <u>loss of cytokeratin-10</u> expression in analysed epithelium,what <u>lower the risk of recurrence</u>
	<ul style="list-style-type: none"> ◆ <u>interleukin-1 (IL-1)</u> stimulates <u>epithelial cell proliferation</u> directly and/or indirectly by inducing the secretion of keratinocyte growth factor from interacting fibroblasts. ◆ The proliferating activity of the epithelial cells is related to the expansion of KCOT. Therefore, <u>IL-1A</u> is considered to be an <u>important factor in regulating the KCOT growth</u>.
Ninomiya et al.	<ul style="list-style-type: none"> ◆ the epithelial cells of KCOT express IL-1A mRNA and protein →and <u>decompression by marsupialization</u> immensely <u>reduce the expression proportionally with the epithelial cell proliferation</u>. →<u>decompression offers significant advantages in decreasing the size of the tumour</u>.
	<ul style="list-style-type: none"> ◆ The majority of research in this area indicates that <u>most recurrences appear within the first 5–7 years</u> →; however, relapse <u>may occur 9 or more years after the initial</u>

	<p><u>treatment.</u></p> <ul style="list-style-type: none"> ◆ Established results have shown <u>different recurrence rates for specific treatment modalities.</u> ◆ Despite many studies indicative of <u>radical methods advantages</u> over conservative treatment, →<u>no conclusive proof</u> has been established convincing that <u>conservative methods do not increase the success rate in treatment of KCOT.</u>
Marker et al.	<ul style="list-style-type: none"> ◆ reported a recurrence rate of <u>8.7%</u> →after <u>decompression with subsequent enucleation</u> of the tumour
	<ul style="list-style-type: none"> ◆ Despite the fact that <u>decompression requires longer time</u> when compared with other methods of treatment →it <u>reduces the chance of a pathological fracture, loss of teeth vitality, or bony discontinuity</u> with definitive treatment. ◆ It needs to be emphasised that <u>systematic and long-term follow-ups are considered to be a key element</u> for successful results.
題號	題目
1	<p>下列關於顎骨中囊腫之臨床表徵描述，那些錯誤？①除非繼發感染，很少在早期出現症狀 ②很少因壓迫神經而造成感覺異常 ③常造成鄰近牙根的吸收</p> <p>(A) 只有① (B) 只有①② (C) 只有③ (D) ①②③</p>
答案(C)	出處：Oral and Maxillofacial Pathology, Nevielle, P131
題號	題目
2	<p>對於正常角質化齒源性角化囊腫（orthokeratinized odontogenic keratocyst）的敘述，下列何者錯誤？</p> <p>(A) 在X光片下，大多是呈現單房性（unilocular）的放射性透光病灶 (B) 與含齒囊腫（dentigerous cyst）相似，也都會有伴隨著未萌發齒 (C) 復發率極高 (D) 挖出術（enucleation）加刮除術（curettage）是很好的治療選擇</p>
答案(C)	出處：Oral and Maxillofacial Pathology(2nd), P.59-8