# B-ENT, 2017, 13, 321-326

# Prelacrimal approach for paediatric keratocystic odontogenic tumours involving the maxillary sinus

### H. Kodama<sup>1</sup>, A. Kuboki<sup>1</sup>, K. Hayashi<sup>2</sup>, H. Kojima<sup>1</sup>, N. Otori<sup>1</sup>

<sup>1</sup>Department of Otorhinolaryngology; <sup>2</sup>Department of Dentistry, The Jikei University School of Medicine, Tokyo, Japan

Key-words. Keratocystic odontogenic tumour, pre-lacrimal approach, maxillary sinus, child, endoscope

Abstract. Prelacrimal approach for paediatric keratocystic odontogenic tumours involving the maxillary sinus. Problem: An eight-year-old and a 14-year-old, both male, presented with a keratocystic odontogenic tumour (KCOT) involving the maxillary sinus.

*Methodology:* The KCOT was treated by the prelacrimal approach, which reportedly provides better and less invasive access to the maxillary sinus.

*Results:* In paediatric cases, treatment should prevent nasal structure and permanent tooth damage, while accomplishing total tumour resection. In the presented two cases, the KCOT was successfully treated by the prelacrimal approach without recurrence.

*Conclusions:* The prelacrimal approach is a useful technique in the treatment of paediatric KCOTs involving maxillary lesions. We also discuss the treatment strategies for this disease, along with the efficacy of the prelacrimal approach.

### Introduction

۲

An odontogenic keratocyst (OKC) was originally classified as an odontogenic cyst and known to exhibit a high recurrent rate. Given its aggressive behaviour and frequent recurrence, an OKC with parakeratosis was reclassified as a benign tumour known as a KCOT by the World Health Organization in 2005.1 Previous studies have reported that KCOTs rarely show malignant transformation.<sup>2</sup> For OKCs without keratinization, such as radicular cysts and dentigerous cysts, marsupialization is the only adequate treatment modality.3 However, complete removal has been recommended for KCOTs.24 It is evident that OKCs and tumours require different treatment approaches, despite having the same origin. Therefore, an accurate preoperative diagnosis, which clearly differentiates a cyst from a tumour, is essential in order to determine the optimal treatment approach.

KCOTs and odontogenic cysts often develop in children and occasionally involve the maxillary sinus. For young patients, less invasive surgery is desirable, with due consideration to further growth of the permanent teeth and jaw bone. Previously, several treatment modalities were proposed for KCOTs mainly by dentists or oral surgeons. All these modalities, including total resection, marsupialization and enucleation, required an intraoral approach.<sup>2,4</sup> However, these techniques carry a potential risk of affecting the growth of permanent teeth in children, given that most involve removal of the alveolar bone. Therefore, an alternative approach for complete removal in a less invasive manner is desirable in order to prevent recurrence and unnecessary damage to teeth and bones.

Of late, the indications for endoscopic sinus surgery (ESS) have been expanding, while otolaryngologists have been able to treat various lesions, involving the maxillary sinus, particularly in the posterior region, in a less invasive manner. ESS for maxillary KCOTs and odontogenic cysts involving the maxillary sinus has been performed in a few cases.<sup>5,6</sup> However, conventional ESS does not provide adequate access to the maxillary sinus because of a limited visual field and working space in the anterior region. The prelacrimal

Financial declaration. There are no financial conflicts of interest. This case report was presented at the 54nd Annual Meeting of the Japanese Rhinologic Society in Hiroshima, Japan, October 2, 2015.

### 322

approach is a useful endoscopic approach for maxillary lesions involving the maxillary sinus.<sup>7</sup> This technique provides excellent operability in the maxillary sinus under endoscopic guidance and allows for preservation of the nasolacrimal duct, as well as almost the entire inferior turbinate. Although several recent reports have shown the usefulness of the prelacrimal approach for various maxillary lesions,<sup>89</sup> few studies have reported its use for the treatment of maxillary KCOTs with sinus involvement. To the best of our knowledge, this is the first report referring to the utility of the prelacrimal approach for paediatric KCOTs.

Here, we report on two cases involving an eight-year-old boy and a 14-year-old boy with a KCOT involving the maxillary sinus, which was successfully treated by the prelacrimal approach in both cases, with good midterm outcomes.

# **Case reports**

## Case 1

Case 1 involves an eight-year-old boy who presented with right cheek pain and right maxillary gingival swelling. The patient had undergone surgical drainage of the gingival swelling at a local dental clinic and was diagnosed with a maxillary cyst after radiographic examination. He was referred to our hospital for further investigation and treatment.

Computed tomography (CT) (Figure 1A-C) and magnetic resonance imaging (MRI) (Figure 1D) revealed a massive right-sided cystic lesion occupying the maxillary sinus and containing a tooth. A part of the basal wall was observed to be in close proximity to a permanent tooth germ.

A dentigerous cyst, an ameloblastoma and a KCOT were considered as differential diagnoses before treatment. We consulted with the department of dentistry about the diagnosis and treatment modalities, as well as performed fine-needle aspiration cytology. However, the results were inconclusive because most of the sample included intracystic content, such as macrophages and neutrophils.

The prelacrimal approach was performed to preserve the nasal structures and obtain sufficient manipulation, together with visualization in the maxillary sinus. We placed a mucosal incision vertically at the anterior end of the inferior turbinate. The mucosa was exfoliated from the



H. Kodama et al.

Figure 1

Preoperative CT (A-C) and MRI (D) findings for Case 1 (eight-year-old boy who presented with right cheek pain and right maxillary gingival swelling). A massive cystic lesion occupying the right maxillary sinus and containing a tooth can be observed. Part of the basal wall is close to a permanent tooth germ (arrow).



#### Figure 2

Intraoperative (A-C) and histopathological (D) findings for Case 1 (eight-year-old boy who presented with right check pain and right maxillary gingival swelling). The nasolacrimal duct (\*) has shifted medially with the inferior turbinate, while the anterior part of the cyst wall (star) can be observed after removal of the medial wall of the maxillary sinus (A). A tooth (white arrow) in the tumour wall (dotted line) and a tooth germ (white arrow head) beneath the tumour can be observed (B). Part of the basal wall (broken line) of the tumour is not removed to preserve the adjacent permanent tooth germ (C). Haematoxylin and eosin staining (D) show a parakeratinized epithelium lining the wavy surface of the cystic lumen (black arrow). The basal layer shows palisading columnar cells (black arrow head).

inferior turbinate bone to expose the conchal crest. Then the conchal crest was removed to separate and medialize the inferior turbinate from the

26-Kodama.indd 322

Paediatric KCOT involving the maxillary sinus



Figure 3

Postoperative CT findings for Case 1 (eight-year-old boy who presented with right cheek pain and right maxillary gingival swelling). The permanent tooth germ observed beneath the cyst has erupted (arrow) (A). There is residual cyst tissue (arrow head) without obvious recurrence (B).

lateral nasal wall until the nasolacrimal duct was identified. Subsequently, the nasolacrimal duct was shifted medially with the inferior turbinate and the mucosa of the inferior meatus. Then, the medial wall of the maxillary sinus was exposed and shaved with a bur to enter the sinus cavity. Once the cyst was visualized (Figure 2A), we perforated its wall to decrease its volume and separated the cyst from the wall of the maxillary sinus under the guidance of  $0^{\circ}$  and  $70^{\circ}$  endoscopes (Karl Storz, Tuttlingen, Germany) (Figure 2B). However, following advice from a dentist, part of the basal wall of the cyst was left behind to preserve the adjacent permanent tooth germ (Figure 2C). The resected portion of the cyst was extracted through the nostril.

A histopathological examination with haematoxylin and eosin staining (Figure 2D) of the resected specimen showed a parakeratinized epithelium lining the cystic lumen and a basal layer with palisading columnar cells. The parakeratin layer was observed as a wavy surface. These features typically suggested a KCOT.<sup>1,2</sup>

At one year and three months after surgery, no obvious recurrence was observed via CT. The preserved permanent tooth germ had erupted (Figure 3A). However, careful follow-up will be necessary because of the residual tissue left at the time of surgery (Figure 3B).

# Case 2

Case 2 involved a 14-year-old boy who presented with right cheek swelling and right-sided nasal obstruction. The patient was diagnosed with right maxillary sinusitis and had undergone sinus irrigation at a local ENT clinic. However, as his



Figure 4

Preoperative CT (A, B), MRI (C) and histopathological findings (D) for Case 2 (14-year-old boy who presented with right cheek swelling and right-sided nasal obstruction). A massive cyst lesion occupying the right maxillary sinus and containing a tooth can be observed. There are no adjacent tooth germs. A parakeratinized epithelium (arrow) and a basal layer of palisading columnar cells (arrow head) can be observed.

symptoms did not alleviate, he visited our hospital for further investigation.

Preoperative CT (Figure 4A-B) and MRI (Figure 4C-D) revealed findings similar to those observed for Case 1. A massive cyst containing a tooth was observed in the right maxillary sinus. However, this patient did not have any permanent tooth germs. Similar to Case 1, a dentigerous cyst, an ameloblastoma and a KCOT were considered as differential diagnoses before treatment. The prelacrimal approach was performed by the same surgeon who treated Case 1, with nearly the same

# H. Kodama et al.

324



Figure 5

Postoperative CT findings for Case 2 (14-year-old boy who presented with right cheek swelling and right-sided nasal obstruction)

The tumour was completely removed and there are no signs of obvious recurrence.

procedure. The only difference was that the lack of permanent tooth germs facilitated total resection. The specimen showed the same histopathological findings observed for Case 1, including a parakeratinized epithelium, a palisading basal layer and a wavy epithelial surface. A final diagnosis of a KCOT was made (Figure 5).

No exacerbation of symptoms and local recurrence were observed on the follow-up CT at 10 months after surgery (Figure 5).

### Discussion

۲

We reported two paediatric cases of a KCOT involving the maxillary sinus, which was successfully treated by the prelacrimal approach. KCOTs present with a parakeratinized epithelium lining the cystic lumen, which is histopathologically characterized by a palisading basal cell layer and a corrugated surface.<sup>12,10</sup> Although histopathology is the gold standard and mandatory for a definitive diagnosis, a preoperative biopsy of the maxillary lesion cannot be always performed for paediatric patients under a local anaesthesia because of localization or other factors. In such cases, FNA cytology can be a useful tool in terms of diagnostic assistance. The liquid of a cyst lumen often includes creamy or yellowish keratin debris,<sup>11</sup> which is an important preoperative finding that raises the suspicion of a KCOT. That said, we should be aware that a result with no observation of keratin debris does not necessary exclude the possibility of a KCOT such as in our Case 1.

In terms of other preoperative assessments for a KCOT, some authors have reported the diagnostic usefulness of MRI,<sup>12,13</sup> although diagnosis is generally difficult on the basis of preoperative images alone. Probst et al. described that KCOTs show homogeneous or heterogeneous low-signal intensity in the cystic wall on contrast-enhanced MRI, while odontogenic tumours basically present with homogeneous high-signal intensity.<sup>12</sup> Although preoperative CT, non-enhanced MRI and fineneedle aspiration cytology could not distinguish between a KCOT and other odontogenic cvsts in both our patients, contrast-enhanced MRI may have been helpful for preoperative assessments. However, for patients such as ours, where the possibility of a KCOT cannot be completely ruled out, the treatment plan should be formulated by keeping a KCOT in mind.

The recurrence rate for a KCOT is high. According to previous studies conducted by dentists, the recurrence rate ranges widely from 0% to 62%,<sup>4,10</sup> which implies the lack of a standard established treatment. Younger age, the site of involvement, a large size, a long anteroposterior lesion dimension and multilocularity have been proposed as risk factors for recurrence, while Gonzalez-Alva et al. mentioned that maxillary tumours are at a higher risk of recurrence compared with tumours at other sites.<sup>10,14</sup>Generally, recurrence occurs within 10 years after initial surgery, although recurrence at 20 years after treatment has also been reported.15 This suggests that long-term follow-up is essential, particularly for patients with risk factors for recurrence, such as ours. One of the main causes of recurrence is incomplete removal.<sup>2,4,14</sup> Therefore, complete removal of a KCOT should be mandatory.

However, complete removal of KCOT may not be possible in some paediatric patients. Patients in the mixed dentition stage, which generally lasts until 12 years of age, require tumour removal procedures that ensure the preservation of permanent tooth germs in the vicinity. In our Case 1, who had mixed dentition, it was very difficult to remove the tumour

# Paediatric KCOT involving the maxillary sinus

entirely without damaging a permanent tooth germ that was close to the tumour wall. Therefore, we left behind part of the basal wall in close proximity to the tooth germ (Figure 6A), which ensured normal eruption of that tooth after surgery. This patient needs to be followed up closely for any signs of recurrence and it may be desirable that a yearly CT or MRI is performed at least until 10 years after initial surgery to evaluate the size of the tumour's wall. If the lesion reoccurs, we will perform total resection because the permanent tooth eruption would probably be complete by then. The tumour in Case 2 was completely removed because eruption was complete (Figure 6B). Our findings indicate that it is crucial to plan surgery after considering the status of the permanent dentition in consultation with a dentist.

The prelacrimal approach appears to be an ideal option for paediatric cases of KCOTs involving most of the maxillary sinus. Although several recent studies have reported the effectiveness of an endoscopic approach for odontogenic maxillary lesions, the reported techniques involved partial resection of the cyst wall or staged surgery.6 On the other hand, the prelacrimal approach has been described as a procedure that allows complete resection at the first attempt.8 In both our patients, the prelacrimal approach facilitated access to the entire lesion occupying most of the maxillary sinus, with good operability, no postoperative complications, and preservation of the nasolacrimal duct and inferior turbinate. We believe that the prelacrimal approach has several advantages for paediatric cases with bulky maxillary lesions. However, further research is needed for the assessment of a larger number of patients and longer follow-up periods.

### Conclusions

In conclusion, we reported successful outcomes of the prelacrimal approach for a KCOT involving the maxillary sinus in both a patient in the mixed dentition stage and a patient with completed eruption. The prelacrimal approach is a less invasive procedure, which enables complete tumour removal in most children. When the patient is in the mixed dentition period, the site of the permanent tumour germ should be preserved by incomplete tumour removal. Our results suggest that the prelacrimal approach provides good midterm postoperative

### References

- Philipsen HP. Keratocystic odontogenic tumour. In: Barnes L, Eveson JW, Reichart P, Sidransky D, Eds. *Phatology Genetics of Head Neck Tumors*. IARC Press, Lyon, 2005:306-307.
- 2.Bell RB, Dierks EJ. Treatment options for the recurrent odontogenic keratocyst. *Oral Maxillofac Surg Clin North Am*. 2003;15(3):429-446.
- 3.Manor E, Kachko L, Puterman MB, Szabo G, Bodner L. Cystic lesions of the jaws - a clinicopathological study of 322 cases and review of the literature. *Int J Med Sci*. 2012;9(1):20-26.
- 4.Habibi A, Saghravanian N, Habibi M, Mellati E, Habibi M. Keratocystic odontogenic tumor: a 10-year retrospective study of 83 cases in an Iranian population. *J Oral Sci.* 2007;49(3):229-235.
- 5.Micozkadioglu SD, Erkan AN. Endoscopic removal of a maxillary dentigerous cyst. *B-ENT*. 2007;3(4):213-216.
- 6.Seno S, Ogawa T, Shibayama M, Ogawa F, Fukui J, Owaki S, Suzuki M, Shimizu T. Endoscopic sinus surgery for the odontogenic maxillary cysts. *Rhinology*. 2009;47(3):305-309.
- 7.Weber RK, Hosemann W. Comprehensive review on endonasal endoscopic sinus surgery. *GMS Curr Top Otorhinolaryngol Head Neck Surg*. 2015;14:1-108.
- 8.Nakayama T, Asaka D, Okushi T, Yoshikawa M, Moriyama H, Otori N. Endoscopic medial maxillectomy with preservation of inferior turbinate and nasolacrimal duct. *Am J Rhinol Allergy*. 2012;26(5):405-408.
- 9.Guo T, Sun JW, Wang YF, Sun JQ. Endoscopic endonasal surgery for pterygopalatine fossa schwannoma via prelacrimal recess-maxillary sinus. *B-ENT*. 2014;10(1):81-86.
- González-Alva P, Tanaka A, Oku Y, Yoshizawa D, Itoh S, Sakashita H, Ide F, Tajima Y, Kusama K. Keratocystic odontogenic tumor: a retrospective study of 183 cases. J Oral Sci. 2008;50(2):205-212.
- 11.Bharqava D, Deshpande A, Poqrel MA. Keratocystic odontogenic tumour (KCOT) — a cyst to a tumour. Oral Maxillofac Surg. 2012;16(2):163-170.
- 12. Probst FA, Probst M, Pautke C, Kaltsi E, Otto S, Schiel S, Troeltzsch M, Ehrenfeld M, Cornelius CP, Müller-Lisse UG. Magnetic resonance imaging: a useful tool to distinguish between keratocystic odontogenic tumours and odontogenic cysts. *Br J Oral Maxillofac Surg.* 2015;53(3):217-222.
- Konouchi H, Asaumi J, Yanagi Y, Hisatomi M, Kawai N, Matsuzaki H, Kishi K. Usefulness of contrast enhanced-MRI in the diagnosis of unicystic ameloblastoma. *Oral Oncol.* 2006;42(5):481-486.
- 14. Leung YY, Lau SL, Tsoi KYY, Ma HL, Ng CL. Results of the treatment of keratocystic odontogenic tumours using enucleation and treatment of the residual bony

۲

H. Kodama et al.

326

defect with Carnoy's solution. *Int J Oral Maxillofac Surg*. 2016;45(9):1154-1158.

 Lam KY, Chan C. Odontogenic keratocysts: a clinicopathological study in Hong Kong Chinese. *Laryngoscope*. 2000;110(8):1328-1332. Akihito Kuboki, MD Department of Otorhinolaryngology The Jikei University School of Medicine 3-25-8 Nishi-Shimbashi, Minato-Ku, Tokyo 105-8461, Japan Tel.: +81-3-3433-1111 Fax: +81-3-3578-9208 E-mail: kuboxile@jikei.ac.jp

۲