

Case Report**Modified transnasal endoscopic medial maxillectomy
with inferior turbinate flap for dentigerous cyst*****Retno Sulisty Wardani, *Michael Lekatompessy, **Brent Anthony Senior*****Otorhinolaryngology Head and Neck Surgery Department****Faculty of Medicine Universitas Indonesia/Dr. Cipto Mangunkusumo Hospital
Jakarta******University of North Carolina at Chapel Hill, United States of America****ABSTRACT**

Background: Dentigerous cyst is one of the most frequent types of odontogenic cyst that usually involving an impacted, supernumerary or ectopically erupted tooth. One of the non-dental sites for ectopic eruption is the maxillary sinus. The traditional approach under such circumstances is a Caldwell-Luc maxillotomy, but this type of procedure may result in significant long-term complications. Endoscopic transnasal medial maxillectomy (ETMM) has several advantages, such as good illumination, as well as clear and magnified visualization. The new modified endoscopic transnasal medial maxillectomy (METMM) can provide good visualization and more functional result by preserving the nasolacrimal duct and the inferior turbinate. **Purpose:** We present this case to introduce the METMM technique for extirpation of any tumor in the maxillary sinus. **Case:** One case of dentigerous cyst with an ectopic left maxillary 3rd molar tooth in a 27 year old woman who presented with sinusitis. **Management:** Surgery with a METMM technique to enucleate the cyst, combined with functional endoscopic sinus surgery (FESS) for the sinusitis. The patient then evaluated subjectively for epiphora and objectively with nasoendoscopic examination. **Conclusion:** In this case, METMM was effective in accessing the maxillary sinus allowing for tumor extirpation, while preserving the function of the inferior tubinate and nasolacrimal duct.

Keywords : Dentigerous cyst, maxillary ectopic tooth eruption, transnasal medial maxillectomy

ABSTRAK

Latar belakang: Kista dentigerous merupakan salah satu jenis kista otontogenik yang paling sering ditemukan, biasanya berhubungan dengan gigi impaksi, supernumeri atau gigi yang tumbuh ektopik. Salah satu tempat erupsi ektopik adalah sinus maksilaris. Pendekatan tradisional dalam keadaan ini adalah operasi Caldwell-Luc, tetapi teknik operasi ini dapat mengakibatkan komplikasi jangka panjang. Maksilektomi medial transnasal dengan endoskopi (MMTE) memiliki beberapa keunggulan, seperti visualisasi yang jelas dan diperbesar. Teknik maksilektomi medial transnasal dengan endoskopi yang dimodifikasi (MMTEM) dapat memberikan visualisasi yang baik dan hasil lebih fungsional dengan mempertahankan duktus nasolakrimal dan konka inferior. **Tujuan:** makalah ini diajukan untuk memperkenalkan teknik MMTEM untuk ekstripasi massa yang berada di dalam sinus maksila, karena teknik ini memberikan hasil yang lebih fungsional. **Kasus:** melaporkan satu kasus kista dentigerous dengan gigi molar 3 yang erupsi ektopik pada sinus maksila kiri pada wanita 27 tahun. **Penatalaksanaan:** kasus ini ditatalaksana dengan teknik MMTEM untuk mengenukleasi kista dan BSEF untuk sinusitis. Pasien kemudian dievaluasi secara subjektif dengan anamnesis mengenai adanya epifora dan objektif dengan pemeriksaan nasoendoskopi. **Kesimpulan:** MMTEM terbukti efektif untuk ekstripasi tumor yang berada di dalam sinus maksila, dan teknik ini juga memberikan hasil yang lebih fungsional dengan dipertahankannya konka inferior dan duktus nasolakrimal.

Kata kunci : Kista dentigerous, erupsi gigi ektopik di maksila, transnasal maksilektomi medial

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INTRODUCTION

Dentigerous cysts (DCs) are the most common developmental odontogenic cysts and are derived from the epithelial remnants of the tooth-forming organ. DCs originate by separation of the follicle from around the crown of an unerupted tooth. The mandibular third molar is most often involved, but it may also be associated with an impacted, supernumerary or an ectopically erupted tooth, frequently involving the third molar. Rarely, a tooth or root may be present within the sinus cavity, and such a tooth may have a DC around it. An ectopically erupted tooth associated with a DC in the maxillary sinus may lead to maxillary sinusitis.¹⁻³

The diagnosis of jaw cysts is usually made by clinical symptoms and radiologic findings.¹⁻⁴ Radiologic tests are important in diagnosing cysts that are hard to find with physical examination only and are necessary for planning how the cysts will be treated. With x-ray, DC appear as a distinct radiolucent site with a sclerotic bony border associated with the crown of an unerupted tooth, with the tooth root protruding into the cyst. DCs are mostly unilocular, symmetrically covering the periodontal area. At times, it can enlarge to a size resulting in obstruction of the maxillary sinus. Generally, panoramic radiography is sufficient to diagnose DCs, however, in the case of big cysts, MRI or CT scan may also be necessary.⁵⁻⁷

On MRI, the calcified tooth may not be seen or may be hypodense while the fluid inside the cyst may appear hypodense on T1-weighted images and hyperdense on T2-weighted images. CT scan is useful in deciding the extent of bony destruction.

In cases of pterygoid plate or orbital bone destruction, malignancy should be ruled out.⁸

Histologically, dentigerous cysts have 2-4 cell layers of nonkeratinizing stratified squamous epithelium while keratin elements are rarely found in the inner wall of the cyst. Goblet cells take up 25-50% of all cells that cover the inner wall of the cyst. Other cells such as ciliated epithelial cells, cuboidal cells, columnar cells, hyaline bodies or sebaceous elements are rarely detected. Scattered inflammatory cells can also be found. Hwang⁸ et al reported that the inner wall of the cysts in all cases were covered with stratified squamous epithelium and cuboidal cells, ciliated columnar cells and goblet cells. On rare occasions, squamous cell carcinoma, mucoepidermoid carcinoma or ameloblastoma can develop in dentigerous cysts.

The traditional approach to remove of a DC involves a Caldwell-Luc type of surgery, comprising a sublabial incision followed by an osteotomy of the anterior maxillary sinus wall accompanied by creation of an inferior meatal antrostomy. Although high success rates are reported, this kind of surgery will result in destruction of the function of the sinus and may ultimately result in significant long-term complications, such as facial numbness, sclerosis of the antral walls with collapse of the sinus and subsequent chronic nasal crusting, as well as development of maxillary mucoceles.⁴

Recent technological advances in field of endoscopy have resulted in substantial improvement in techniques of endoscope-controlled surgery of the nose and paranasal sinuses. These techniques has proven very

useful in surgery of the sinuses with its complex anatomy and access limitation. In particular, transnasal endoscopic surgery of the maxillary sinuses for maxillary sinusitis has evidence of good long-term results.⁴

Modified endoscopic transnasal medial maxillectomy (METMM) is a novel procedure wherein under endoscopic visualization, the inferior turbinate and nasolacrimal duct are preserved while the medial maxillary sinus wall is otherwise removed thereby providing access for a complete resection of lesion in the maxillary sinus.⁹⁻¹³ This is accomplished with an incision made in the mucosa of the lateral nasal wall along the anterior margin of the inferior turbinate. The medial maxillary wall is removed while the inferior turbinate and the nasolacrimal duct are preserved and pushed medially allowing wider access into the maxillary sinus. The lesion is removed together with the attachment through a corridor anterior to the nasolacrimal duct. This new surgical technique preserves the whole length of the nasolacrimal unit, reducing the possibility of epiphora, while providing good visualization and no risk of facial numbness.⁹⁻¹³

CASE REPORT

A 25 year old female presented with a chief complaint of intermittent nasal obstruction bilaterally, for two years. There was mucopurulent discharge with foul odor for 5 months, accompanied by post nasal drip, but no history of sneezing, allergies, smell disturbance, nor epistaxis. There was also no history of dental problems.

Nasoendoscopic examination revealed the left lateral nasal wall was pushed medially obstructing the nasal cavity and limiting examination of the posterior sinus and nasal anatomy on that side. Mucopurulent secretions were noted coming out of the middle meatus, while the inferior and middle turbinate were within normal limits.

Dentigerous cyst was suspected from computed tomographic examination, where it was seen to expand the left maxillary sinus thereby obstructing the left nasal cavity and its osteomeatal complex. Opacifications were found in the adjacent left frontal and ethmoid sinuses. A retention cyst was found in the right maxillary sinus.

The patient was diagnosed with a dentigerous cyst of the left maxillary sinus, along with fronto-ethmoid sinusitis, and a retention cyst of the right maxillary sinus.

Dentigerous cyst extirpation by a METMM approach preserving the inferior turbinate and nasolacrimal duct was performed under general anaesthesia. Cotton pledgets soaked in 1/5000 adrenaline/lidocaine solution were inserted for 10 minutes preoperatively. An incision was made at the anterior attachment of inferior turbinate with monopolar cauter until the inferior turbinate bone was exposed, and then the inferior turbinate mucosa was dissected back to the attachment of uncinat process. The uncinat process was removed to expose all the inferior turbinate bone. Elevation of the lateral nasal wall was continued superiorly exposing all of the medial maxillary sinus. The inferior attachment of the inferior turbinate was cut with the monopolar cautery until its posterior attachment, then the lateral nasal wall mucosa and inferior turbinate were shifted medially. Using a sharp chisel, all of the inferior turbinate bone was removed while preserving its posterior attachment. The lacrimal bone and the nasolacrimal duct were identified carefully and the lacrimal bone was removed in order to shift the nasolacrimal duct medially. The opening of the maxillary sinus was widened superiorly to expose the roof of the sinus. The dentigerous cyst was then enucleated along with an ectopic 3rd molar, while the bony part (hyperostosis) that surrounded the cyst was trimmed using a bone cutter and the attachment was cauterized using a

monopolar cauter. The polypoid maxillary mucosa around the cyst was also removed. The natural maxillary ostium was widened postero-inferiorly.

The bulla ethmoid and the basal lamella mucosa were polypoid. The bulla was removed and polypoid mucosa trimmed with a through-cutting forceps. The bleeding was controlled with adrenaline/lidocaine soaked cotton pledgets. The lateral nasal mucosa and inferior turbinate were then returned to its original position. The retention cyst in the right maxillary sinus was removed using the maxillary sinus forceps after uncinectomy and middle meatal antrostomy. Both of the nasal cavities were packed.

The patient was hospitalized and the nasal packing was removed the next day. There was some blood clot and mucoid discharge but no active bleeding. The anterior part of the inferior turbinate was not attached yet to the lateral nasal wall so gelfoam was put at

the attachment to create a soft tissue bridge. The patient was discharged and instructed to take oral antibiotic, analgesic, and to do nasal irrigations with normal saline.

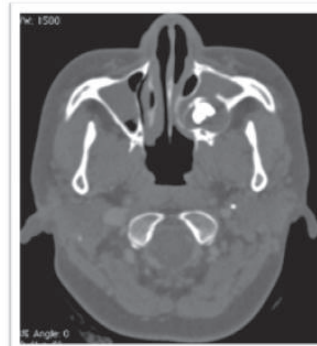
At one week follow-up, the anterior part of the inferior turbinate was found attached to the lateral nasal wall. The left maxillary sinus was widely open.

The pathology examination revealed tissue that resembled a cyst wall which contained ciliated pseudostratified columnar epithelial cells with goblet cells. The connective tissue stroma was filled with chronic inflammatory cells and blood vessels. There were no signs of malignancy.

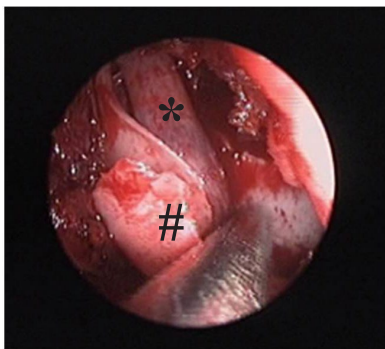
Two weeks postoperative, the patient did not have any complaint of epiphora. The physical examination showed an intact left lateral nasal wall, healthy inferior turbinate, and wide openings into both maxillary sinuses.



Picture 1. CT scan imaging of dentigerous cyst coronal view



Picture 2. Axial view



Picture 3. Before lacrimal bone removal

* Nasolacrimal duct
Lacrimal bone



Picture 4. After lacrimal bone removal



Picture 5. Incision line above inferior turbinate

EVIDENCE BASED SURGERY ANALYSIS

Clinical Question

Is the endoscopic approach better for the treatment of a dentigerous cyst in the maxillary sinus compared to the Caldwell-Luc approach?

Literature Search

On June 4, 2013 and June 17, 2015 a literature search was done using PUBMED, Highwire, Sciencedirect, with the keyword “endoscopic” AND “dentigerous cyst” and we obtained 55 citations. Literature search was continued by changing the keyword to “endoscopic” AND “dentigerous cyst” AND “Caldwell Luc”, and we obtained 14 citations.

From the 14 citations only 6 citations met the inclusion and exclusion criteria, but none of them had the highest evidence which is the RCT (Randomized Controlled Trial) or meta analysis, or any study that compares these procedures. The inclusion criteria for analysis is a case report of serial cases of dentigerous cyst underwent endoscopic trans-nasal surgery. Modified trans-nasal medial maxillectomy for pathology other than dentigerous cyst were excluded.

Xu et al¹⁴ performed functional surgery for 20 cases of dentigerous cyst in maxillary sinus. Functional surgery by their definition is creating a bony lid anterolaterally of maxillary sinus and titanium plates re-implantation after enucleating the cyst, leaving the maxillary sinus mucosa in place. Although the lesions were completely removed, moderately post-operative facial pain (VAS 4-6 of 10) and swelling in the infra-orbital region still occurred in 15% and 40%. The title and the definition of the procedure described is a Caldwell Luc with replacement of the bone, and it is not fit with a functional surgery. This literature is excluded from the analysis.

Analysis

In this case, the clinical question cannot be answered using evidence-based literature review analysis. A major limitation in cases of surgery is the difficulty of achieving blinding and treatment randomization.

Even though the clinical question can not be answered, a report of endonasal removal of dentigerous cyst in the maxillary sinus by Bae et al¹¹ illustrates several advantages of an endoscopic approach, including good illumination, clear and magnified visualization, resulting in more directed surgery and precise dissection. Postoperative course was uneventful and there was no recurrence at 4 years follow-up.

Seno et al¹⁰ identified several interesting advantages of endoscopic sinus surgery for an odontogenic maxillary cysts. This technique can be applied in cases with odontogenic maxillary cysts since it is less invasive than conventional dental procedures, and it prevents oroantral fistula formation and chronic maxillary sinusitis. there were no complications during or after endoscopic surgery.

A series by Sung et al, ¹¹ reported 13 patients (male 6 and female 7, 19 to 75 years old) with odontogenic maxillary cysts that extended into the maxillary sinus. Ten patients had a radicular cyst and three patients had a dentigerous cyst, all were treated with endoscopic approach. In the three patients with a dentigerous cyst, there were no complications, and postoperative courses were uneventful. In five of ten patients with a radicular cyst, they reported removing the cyst walls completely from the maxillary sinus. Follow-up period ranged from 11 to 72 months (mean 42 months), and no recurrence was noted in any of the cases. Advantages of the endoscopic approach noted by these authors include that it was less invasive than conventional dental procedures, and it prevented oroantral fistula formation and chronic maxillary sinusitis.

Another five patients underwent partial resection of the cyst walls of the radicular cyst. However, no recurrence has been noted, also in the same period of follow-up. These results indicate that the partial resection of the cyst wall may be sufficient for the endoscopic treatment of the radicular cysts. In patients with a DC, the cyst walls was completely removed in all three cases. Because malignant change in the dentigerous walls cysts have been reported, the walls of the dentigerous cysts should be completely removed.¹¹

DISCUSSION

According to the literature dentigerous cysts are usually asymptomatic. However, in cases of enlarged cysts or secondary inflammation of the cyst, symptoms such as facial swelling, sensory changes and fistula formation could occur. Patients with DC over the maxillary sinus might present with symptoms of rhinosinusitis. In addition, ophthalmologic symptoms might be present such as proptosis, diplopia, ptosis, epiphora, however visual acuity is rarely affected.

Panoramic x-ray is the most frequently performed radiologic examination given their dental origin, but in our case CT-scan was performed because there was no specific symptoms, and signs from anamnesis and physical examination leading to maxillary sinus tumor instead of dentigerous cyst. CT examination was useful in deciding the extent of bony destruction and evaluating the extent of sinus pathology.

The pathological findings of dentigerous cysts are generally nonkeratinizing stratified squamous epithelium consisting of 2-4 cell layers while keratin elements are rarely found in the inner wall of the cyst. Mucous producing cells make up 25-50% of all cells that cover the inner wall of the cyst. Other cells such as ciliated cells, cuboidal cells, columnar cells, as well as hyaline bodies or sebaceous elements

are rarely detected. Invasion of inflammatory cells can be found. On rare occasions, squamous cell carcinoma, mucoepidermoid carcinoma or ameloblastoma can develop in dentigerous cysts. In our case, the pathology was consistent with a dentigerous cyst without any sign of malignancy.

Since its introduction, the Caldwell-Luc procedure has become a standard approach for the management of antral disease as well as surgical access for pterygomaxillary space, orbit, ethmoid labyrinth and medial skull base. The development of endoscopic sinus surgery has expanded from use in management of inflammatory disease to tumors and other sinonasal lesions and may be indicated for DC removal. The transnasal endoscopic approach prevents the complications and morbidity of traditional procedures including oroantral fistulas, chronic maxillary sinusitis, loss of dentition and effects on maxillary bone growth. In addition, this approach eliminates the need for extensive maxillary reconstruction that may be required with open maxillectomy techniques.

A new paradigm of doing minimal invasive surgery with the aim to preserve physiologic function, while minimizing morbidity and preventing complications has emerged with the development of various techniques in a variety of surgical disciplines.

Xu et al,¹⁴ who are maxillofacial surgeons preferred functional surgery of modified Caldwell-Luc instead of transnasal endoscopic approach due to narrow field of view, moderate learning curve, and difficult operation for dead angles. On their opinion, trans-nasal endoscopic is not adequate to remove large lesions and cysts that lie medially or contain teeth such as impacted teeth, DCs, and dental roots in the maxillary sinus through a small orifice. This argumentative opinion is a challenge for rhino-surgeon to master the knowledge of lateral nasal wall anatomy and the dexterity of rigid nasal endoscope handling.

The modified endoscopic transnasal medial maxillectomy technique applied in this case was able to preserve the inferior turbinate and nasolacrimal duct and thereby preserving the physiologic function, whereas in the standard endoscopic medial maxillectomy those structures are sacrificed. Even though this technique was first performed for inverted papilloma in the maxillary sinus cases, this case showed that this technique could also be used for DCs and no complications of sinusitis nor epiphora appeared in a 14 months follow-up.

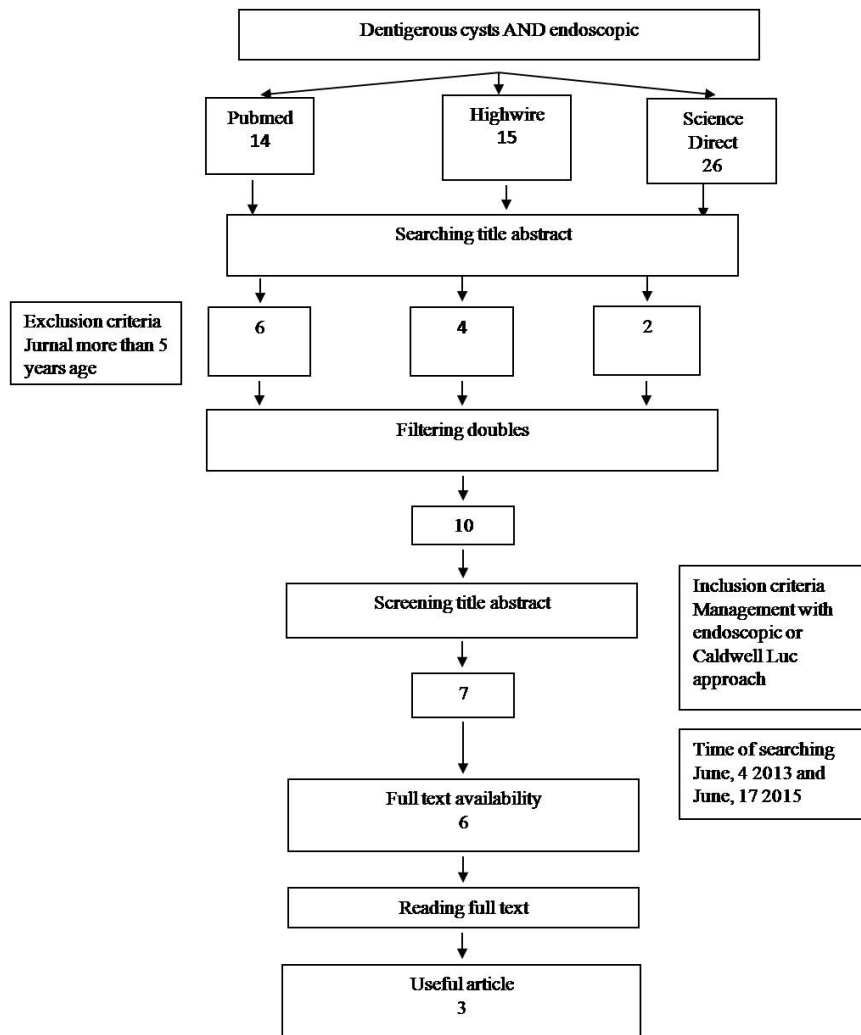
The decision to choose what type of surgery for certain cases is significantly reliant on the familiarity of the surgeon with the lateral nasal wall anatomy and the surgeon's skill level and dexterity. Although there has been no literature review comparing Caldwell-Luc and METMM, we found that the latter procedure gives good results, providing sufficient access to the sinus for good visualization and tumor extirpation, while preserving the inferior turbinate and nasolacrimal duct, with less risk of post-operative complication.

REFERENCE

- Girish G, Umashankar D, Sharma R, Veeresh M, Bhandari A. Dentigerous cyst in maxillary sinus: A rare occurrence. *International Journal of Oral & Maxillofacial Pathology*. 2011;2:20-3.
- Wang P, Wang Y, Shyng Y, Kao W. Dentigerous cyst over maxillary sinus: A case report and literature review. *Taiwan J Oral Maxillofac Surg*. 2009;20:116-24.
- Amin M, Khairudin A. Removal of extensive maxillary dentigerous cyst via a Caldwell-Luc procedure. *Archives of Orofacial Sciences*. 2008;2(3):48-51.
- Andrić M. Endoscopic surgery of maxillary sinuses in oral surgery and implantology. In: Iancu PC, editor. *Advances in Endoscopic Surgery*; InTech; 2011.
- Shankar L ek. An atlas of imaging of the paranasal sinuses. gross and sectional anatomy of the nasal cavity and paranasal sinuses: crc press; 2006. p. 12.
- Gabriele R, Gaudy J, Fabbro M. Maxillary sinus vascularization: A cadaveric study. *The Journal of Craniofacial Surgery*. May 2009; Volume 20(3).
- Pradeep K, Mohapatra N. Conservative management of a dentigerous cyst associated with an impacted mandibular second premolar in mixed dentition. *JODDD*. 2009;Vol. 3.
- Hwang S, Kim D, Lee D, Lee J, Choi J, Lee S. Dentigerous cyst involving the maxillary sinus. *J Rhinol* 2001;8:54-7.
- Nakamaru Y, Takagi D, Oridate N, Fukuda S. Preservation of the nasolacrimal duct during endoscopic medial maxillectomy for sinonasal inverted papilloma. *Rhinology*. 2010;48:452-6.
- Seno S, Shibayama M, Ogawa F, Fukui J, Owaki S, Suzuki M, Shimizu T. Endoscopic sinus surgery for the odontogenic maxillary cysts. *Rhinology*. 2009;47:305-9.
- Bae S, Mo J, Chung Y. Endonasal removal of dentigerous cyst in the maxillary sinus. *Korean J Otorhinolaryngol-Head Neck Surg*. 2013(56):53-5.
- Suzuki M, Nakayama M, Inagaki A, Murakami S, Takemura K, Yokota M. Modified transnasal endoscopic medial maxillectomy with medial shift of preserved inferior turbinate and nasolacrimal duct. *Laryngoscope*. 2011;121(11):2399-401. Epub 2011 Oct 12.
- Nakayama T, Okushi T, Yoshikawa M, Moriyama H, Otori N. Endoscopic medial maxillectomy with preservation of inferior turbinate and nasolacrimal duct. *American Journal of Rhinology & Allergy*. 2012; 26(4):405-8.
- Xu GC, Yang C, Fan XD, Hu YK, Yu CQ. Functional surgery for the treatment of dentigerous cyst in the maxillary sinus. *J Craniofac Surg*. 2015;26(2):84-6.

Appendix

Literature Search Flowchart



Appendix**Critical Appraisal Evidence Based Surgery**

Criteria	Study			
Validity	Seno, 2009	Bae, 2013	Xu, 2015	Nakamaru, 2010
1. Was patient assignment randomized, and the randomization process “concealed”?	No	No	No	No
2. Were all patients who entered the trial accounted for?	No	No	No	No
3. Was follow-up adequate? Were patients analyzed according to the “intention to treat” principle?	Yes	Yes	Yes	Yes
4. Were study personnel “blinded” to treatment?	No	No	No	No
5. Were the patient groups similar before treatment?	No	No	No	No
6. Apart from the experimental intervention, were the groups treated equally?	No	No	No	No
Importance				
1. How large was the treatment effect?				
2. How precise was the estimate of the treatment effect?				
Applicability				
1. Were the study patients similar to my patients?	Yes	Yes	Yes	Yes
2. Were the measured outcomes clinically relevant?	Yes	Yes	Yes	Yes
3. Are my surgical skills similar to those of the study surgeons?	Yes	Yes	Yes	Yes