CASE REPORT

Surgical ciliated cyst of the mandible after orthognathic surgery: a case report with review of the literature

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Abstract
Background: Surgical ciliated cysts, also known as postoperative maxillary cysts or implantation cysts, occur mainly in the posterior maxilla after radical maxillary sinus surgery; they rarely develop in the mandible. They are thought to occur when the sinonasal epithelium is infiltrated by a surgical instrument during surgery or as a result of transplantation of bone or cartilage with respiratory epithelium attached.

Case presentation: We report a case in which a surgical ciliated cyst developed in the anterior part of the mandible, presumably as a result of bimaxillary orthognathic surgery and genioplasty performed 24 years earlier. We then review the few similar cases reported in the literature.

Conclusion: Surgical ciliated cysts in the mandible are extremely rare, but they could occur after simultaneous surgery on the maxilla and mandible, even decades later. To prevent surgical ciliated cysts in the mandible, we recommend that the surgical instruments, especially the saw blade used during bimaxillary surgery, be new or cleaned and that previously placed plates and screws be removed at an appropriate time.

Keywords: Surgical ciliated cyst, Postoperative maxillary cyst, Implantation cyst, Orthognathic surgery

Background
Surgical ciliated cysts, first reported in 1927 by Kubo, are complications that occur after radical maxillary sinus surgery [1]. They are also known as postoperative maxillary cyst, implantation cyst, ectopic ciliated cyst, and respiratory cyst [2–4]. Most surgical ciliated cysts are found in the maxillary molar area. The reported incidence has been high in Japan, where these lesions account for 20% of oral cysts [5, 6]. They have respiratory-type epithelium lining inside, and the presumed cause is infiltration of the mucous membrane of the maxillary sinus by an instrument during surgery [6]. Surgical ciliated cysts in the mandible are very rare; only 15 cases have been reported in English-language journals so far [2–4, 7, 8]. We report a 16th case of surgical ciliated cyst in the mandible, presumed to have occurred after orthognathic surgery and genioplasty performed 24 years earlier.

Case presentation
A 42-year-old man was referred to the Department of Oral and Maxillofacial Surgery, Seoul National University Dental Hospital, for treatment of a cyst in the anterior mandible that was found at a local dental clinic. Cone-beam computed tomography revealed a radiolucent lesion with a diameter of > 2 cm and a labial cortical bone perforation in the lower anterior region, and the cyst was in contact with two fixed metal plates (Fig. 1). The patient stated that he had undergone orthognathic surgery and genioplasty at another hospital 24 years earlier. The patient had no medical history except for smoking half a pack a day, and he did not complain of any other symptoms such as pain or paresthesia in the affected area.
While the patient was under general anesthesia, the cyst was enucleated. A full-thickness alveolar mucosal flap was constructed via an incision in the anterior part of the mandible. The anterior part of the cyst was attached to the periosteum and was dissected and enucleated (Fig. 2). After resection of the soft tissue remaining on the cyst wall, a specimen was sent to the Department of Oral Pathology for definite diagnosis. Two four-hole plates and eight screws were removed. Primary sutures with absorbable, and nonabsorbable suture thread were placed. Histopathological examination revealed that the cyst was lined inside with ciliated respiratory epithelium, and a surgical ciliated cyst was diagnosed (Fig. 3). No recurrence was observed on panoramic radiographs 7 months after the surgery (Fig. 4).

**Discussion**

Surgical ciliated cysts in the mandible are extremely rare. To the best of our knowledge, only 15 cases other than ours have been reported in English-language journals so far; they are summarized in Table 1. The lesions were discovered 2–56 years after surgery, which was presumed to be the cause of the cysts, and most affected patients had signs and symptoms such as swelling, pain, or pus discharge. Of the 16 cases, 12 occurred in the anterior part of the mandible. In all reported cases, patients had undergone multiple concomitant surgical procedures in the areas where the cysts developed. Most of the anterior mandibular cysts occurred in patients who underwent surgery in the chin region, such as chin augmentation or genioplasty accompanying a septoplasty or LeFort I osteotomy; in contrast, mandibular ramal cysts can occur after bimaxillary orthognathic surgery.
The major difference between surgical ciliated cysts in the maxilla and those in the mandible is the developmental cause. Maxillary surgical ciliated cysts are hypothesized to develop either from the entrapment of sinus mucosal remnants or as a result of early closure of the maxillary ostium. The lesions can develop after sinus surgery; LeFort I, II, or III osteotomy; trauma; or traumatic extraction [16, 17]. Mandibular surgical ciliated cysts, however, are preceded by simultaneous surgery on the maxilla and mandible, such as chin augmentation with septal cartilage or bimaxillary orthognathic surgery. Of the 16 cases reported, 8 developed after nasal cartilage and bone transplantation, which supports the hypothesis that respiratory epithelium attached to the graft and transferred to the surgical site gives rise to surgical ciliated cysts [15].

Conversely, cysts developed in seven patients who had not undergone transplantation, which supports the hypothesis that the sinonasal epithelium attached to the saw blade used for maxillary osteotomy is transferred to the mandible, which gives rise to the cyst [5]. To prevent this, do not include the treatment option of bone or cartilage transplantation at the time of treatment planning, or if a cutting saw is used to cut the maxilla, a new one should be used before mandibular osteotomy [4, 5, 7]. Furthermore, to prevent the sinonasal epithelium from being transferred to the mandible by the surgical instrument, it would be advantageous to clean or replace not only the cutting saw but also other surgical instruments, such as drill bits and chisels. In only 1 of the 16 cases—the one published most recently before our report—a mandibular surgical ciliated cyst was observed after grafting of platelet-rich plasma on the extraction socket, without simultaneous maxilla and mandible surgery [8]. Those authors assumed that the cyst was caused by growth factor in the platelet-rich plasma, but this was the only such case, and more research on the causes is needed in the future.

As in our case, if the cyst is attached to the bony wall and to the soft tissue, complete removal could be difficult, and the lesion may recur. According to Soares et al., of the 17 ciliated cysts resected, none recurred [18]. In that study; however, the follow-up after removal of the lesions was short (mean: 8.6 months), and the number of samples was small; therefore, we recommend regular check-ups after surgery. Also, it may be recommended that any plate and screw used for previous orthognathic surgery could be removed at an appropriate time to reduce the possibility of cyst occurrence.

In summary, surgical ciliated cysts in the mandible are extremely rare, but they could occur after simultaneous surgery on the maxilla and mandible, even decades later. For differential diagnosis, periapical cysts or odontogenic keratocysts, similar to osteolytic, should be ruled out. To prevent surgical ciliated cysts in the mandible, we recommend that the surgical instruments, especially the saw blade used during bimaxillary...
surgery, be new or cleaned and that previously placed plates and screws be removed at an appropriate time. Because little is currently known about recurrence of surgical ciliated cysts, we also recommend long-term follow-up after cyst enucleation.

**Abbreviation**

BSSO: Bilateral sagittal split osteotomy.

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**Authors’ contributions**

SY wrote the manuscript, HO has participated in conceptualizing the report, HY has provided the pathologic image and explanation, and BS has taken the role of initializing and confirming the manuscript. The authors read and approved the final manuscript.

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Availability of data and materials
Data sharing is not applicable to this report as no data sets were generated or analyzed during the current study.

Declarations

Ethics approval and consent to participate
No consent to participate was obtained since the data collected was retrospective and did not include information on personal identification. This case report was approved by the Institutional Review Board (IRB) of the Seoul National University Dental Hospital at Seoul (IRB no. ER12.2016).

Consent for publication
Not applicable, because no personally identifiable data was included in this report.

Competing interests
The authors declare that they have no competing interests.

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References
1. Kubo I (1927) A buccal cyst occurred after a radical operation of the maxillary sinus. Z f Otol Tokyo 33:896
2. Li C-C, Feinerman DM, MacCarthy KD, Woo S-B (2014) Rare mandibular surgical ciliated cysts: report of two new cases. J Oral Maxillofac Surg 72:1736
3. Syed N, Mason R, Thomson E, Downie J (2018) Mandibular respiratory cysts following orthognathic surgery: 2 rare case reports. Int J Surg 55:568
4. Selit S, Sohianian S, Khalkazz D, Abesi F, Alakbarpour F, Rayani A (2016) Ectopic ciliated cyst in the mandible secondary to genioplasty and lefort after two years: a case report and literature review. Iran J Otorhinolaryngol 28:353
5. Bourgeois SL, Nelson BL (2005) Surgical ciliated cyst of the mandible secondary to simultaneous Le Fort I osteotomy and genioplasty: report of case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 100:36
6. Bulut AŞ, Şehlaver C, Perçin AK (2010) Postoperative maxillary cyst: a case report. Patholog Res Int 2010:810835
7. Cai M, Shen G, Lu X, Wang X (2015) Two mandibular surgical ciliated cysts after Le Fort I osteotomy and genioplasty. Br J Oral Maxillofac Surg 53:1040
8. Lafuente-Ibáñez de Mendoza I, Fernández-Reyes M, Fernández-Arenas A, Aguirre-Unzur JM (2021) Surgical ciliated cyst after a mandibular surgery: a particular case report and review of the literature. BMC Oral Health 21:1
9. Naistri AL, Hooke PY (1994) Respiratory epithelium in a mandibular cyst after grafting of autogenous bone. Int J Oral Maxillofac Surg 23:372
10. Anastassov GE, Lee H (1999) Respiratory mucocele formation after augmentation genioplasty with nasal osteocartilaginous graft. J Oral Maxillofac Surg 57:1263
11. Kelly JP, Malik S, Stucky-McCormick SU (2000) Tender swelling of the chin 40 years after genioplasty. J Oral Maxillofac Surg 58:203
12. Imholte Drmeddent M, Schwartz HC (2001) Respiratory implantation cyst of the mandible after chin augmentation: report of case. Otolaryngol Head Neck Surg 124:586
13. Koutlas IG, Gillum RB, Harris MW, Brown BA (2002) Surgical (implantation) cyst of the mandible with ciliated respiratory epithelial lining: a case report. J Oral Maxillofac Surg 60:324
14. Lazar F, zur Hausen A, Mischikowski R, Zöller JE (2006) Atypical cyst formation following chin augmentation using a nasal osteocartilaginous graft. J Craniomaxillofac Surg 34:107
15. Ragsdale BD, St Jessica LL, Janette AJ, Epker BN (2009) Respiratory implantation cyst of the mandible following orthognathic surgery. J Oral Maxillofac Pathol 13:30
16. Gol asbestoski J, Munoz R, Barazarte D, Perez L (2019) Surgical ciliated cyst after maxillary orthognathic surgery: a literature review and case report. Oral Maxillofac Surg 23:281
17. Tanio S, Tamura T, Kasuya H, Kawasaki M, Taniguchi N, Otsuki K, Fujii N, Kodani I (2019) Surgical ciliated cyst developing after Le Fort I osteotomy: case report and review of the literature. J Oral Maxillofac Surg Med Pathol 31:410
18. Soares JC, Villalba NC, Sanromán JF, Ferro MF, Fernández PL, Betancourt AL, López AC (2021) Surgical ciliated cysts in orthognathic surgery. J Craniomaxillofac Surg 32:22

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