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Case report

Surgical ciliated cyst developing after Le Fort I osteotomy: Case report and review of the literature

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ABSTRACT

A surgical ciliated cyst, also referred as a locally aggressive lesion, stems from a prior surgery several years later as a delayed complication. It is reported that the cyst develops after orthognathic surgery. A 25-year-old man who had undergone Le Fort I osteotomy and bilateral intraoral vertical osteotomy of the mandible as treatment for jaw deformities in 2013 was referred to our hospital because of a swelling on the right side of the hard palate. Radiography and cone-beam computed tomography examinations revealed a unilocular cystic lesion in the right maxillary alveolus. All the teeth in the maxillary right quadrant were vital to electric pulp testing. Aspiration with an 18-gauge needle extracted rust-colored mucoid fluid. The lesion was diagnosed to be a maxillary cyst. Under general anesthesia, we removed the osteotomy fixation plates and excised the lesion.

Histopathological examination revealed a cyst comprising cellular fibrous connective tissue, mainly lined by stratified squamous epithelium; however, the regions of focal pseudostratified columnar and ciliated epithelium were also present. Surgical ciliated cyst was diagnosed on the basis of histopathological, clinical, and radiographical findings.

Although surgical ciliated cysts have sparsely been reported after orthognathic surgery, an increased awareness of such possibility is required to avoid delays in diagnosis. This lesion should always be a part of differential diagnosis of symptomatic patients who, in the past, have undergone antral or maxillary orthognathic surgery.

1. Introduction

In 1927, Kubo was the first to report a surgical ciliated cyst as a posterior maxillary cyst after the surgical treatment of maxillary sinusitis [1]. It was presented as a delayed complication following a surgery in the region of the maxillary sinus after Caldwell-Luc radical antrostomy. Although sparsely reported in English journals, numerous reports of the lesion occur in Japanese literature, wherein it is frequently referred to as a "postoperative maxillary" or "paranasal cyst." It is one of the most frequently encountered bony cysts of the jaws and is reported to occur in up to 20% of patients who have undergone a radical surgery on the maxillary sinus [2–4]. In 1990, Sugar, Walker, and Bounds reported three cases of surgical ciliated cyst following midface orthognathic surgery [5]. Here we present the case of a surgical ciliated cyst, which developed within the anterior hard palate 5 years after a segmental Le Fort I osteotomy. To the best of our knowledge, this represents the first reported case of a surgical ciliated cyst that occurred

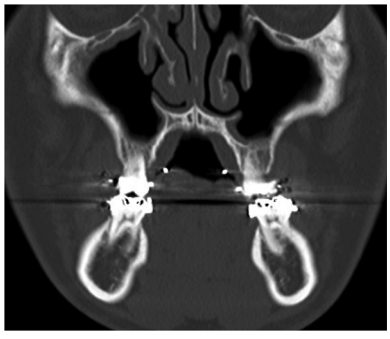

Fig. 1. Coronal computed tomography before orthognathic surgery. There was no cystic lesion in maxillary alveolus.

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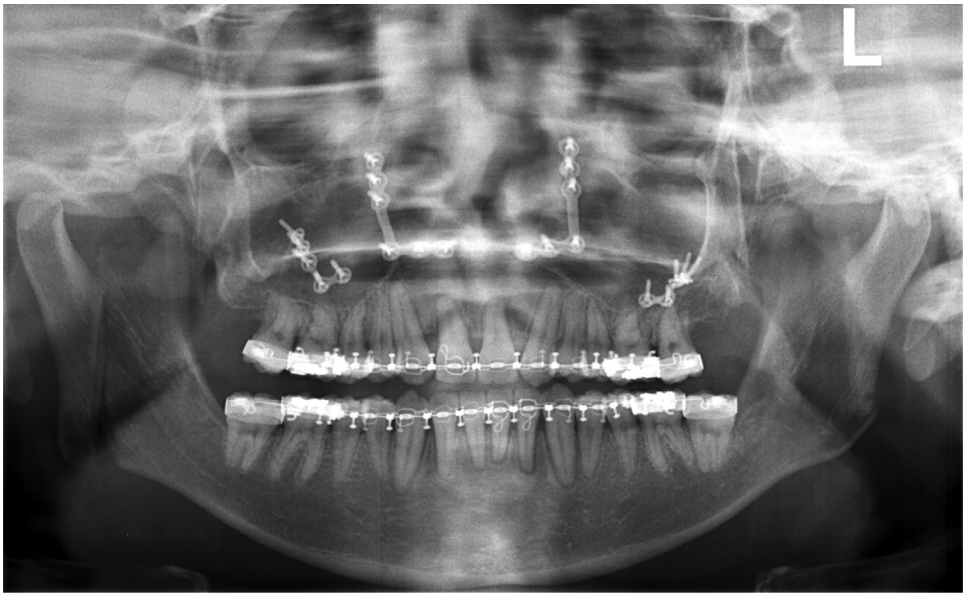

Fig. 2. Panoramic radiograph 1 week postoperatively.

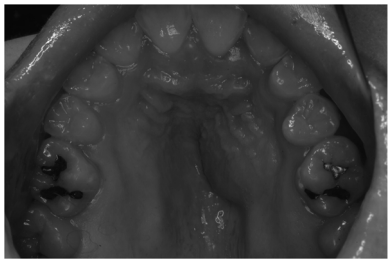

Fig. 3. Intraoral view showing a swelling in the palate (mirror image).

after maxillary orthognathic surgery within the anterior hard palate and in association with nasal mucosa. Misch et al described a case of postoperative maxillary cyst that developed after a sinus elevation procedure in 1991 [6], and Toyoshima et al described a case of traumatic ciliated cyst from zygomaticomaxillary fracture in 2014 [7].

2. Case report

In early-February 2012, a 20-year-old man diagnosed as jaw deformities at orthodontic clinic visited our hospital for the purpose of preoperative examination. There was no problem in the examination result, and he was started preoperative orthodontic treatment (Fig. 1).

In 2013, he underwent a Le Fort I maxillary osteotomy and a bilateral intraoral vertical osteotomy of the mandible as a treatment for jaw deformities at our hospital. No immediate postoperative complications were noted (Fig. 2). The patient's postoperative course was uneventful. He was scheduled for a surgery to remove the osteotomy fixation plates from the bilateral maxilla, although he interrupted to outpatient visit. Early in February 2017, he had a gradual swelling on the right side of the hard palate for which he consulted a family doctor. In late-February 2017, he was referred to our department for more intensive examination and treatment of the swelling, which was the chief complaint.

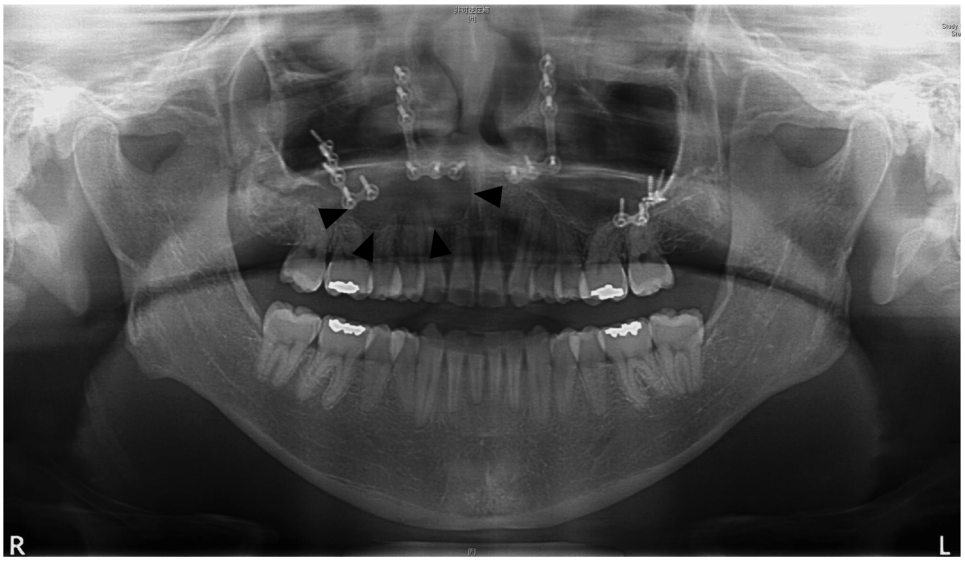

Fig. 4. Panoramic radiograph showing unilocular, radiolucent image (arrowhead) from the region of tooth number 11 to 16.

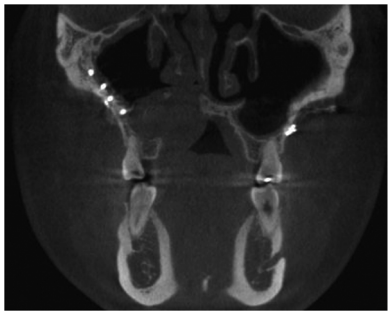

Fig. 5. Coronal cone-beam computed tomography showing an extensive, unilocular cystic lesion in the right maxillary alveolus. The floor of the maxillary sinus had been displaced superiorly.

Intraoral findings revealed a well-defined swelling, which was accompanied by a fluctuation, measuring approximately 15 mm in diameter (Fig.3).

Panoramic radiograph revealed a well-defined, unilocular, radiolucent image from the region of tooth number 11 to 16, and there was no resorption of the root or displacement (Fig. 4). All the teeth in the maxillary right quadrant were vital to electric pulp testing. Cone-beam computed tomography revealed an expansive unilocular lesion in the right maxillary alveolus (Fig. 5). The floor of the maxillary sinus had been displaced superiorly. In addition, a partial bone defect was observed in the labial and palatal side of the maxilla (Fig. 6).

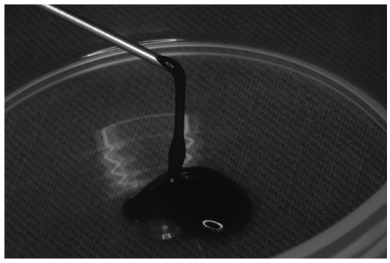

Fig. 7. Aspiration with an 18-gauge needle extracting rust-colored mucoid fluid.

Aspiration with an 18-gauge needle extracted rust-colored mucoid fluid (Fig. 7). Neither odor nor purulent drainage was observed.

After maxillary orthognathic surgery, on the basis of clinical findings, imaging examination, and rust-colored mucoid fluid, a clinical diagnosis of the maxillary cyst on the lesion was made. The patient was scheduled to be operated for the removal of the osteotomy fixation plates and lesion.

Under general anesthesia, the osteotomy fixation plates on the left side maxilla were first removed. Subsequently, a Partsch incision was made from the region of tooth number 11 to 17, and the mucoperiosteal flap was elevated to expose the anterolateral wall of the maxillary sinus. The osteotomy fixation plates on the right side maxilla were removed and the bone was thinned with a perforation in the region of tooth number 13 (Fig. 8). A bone window was created, showing a smooth, thick-walled, and unilocular cyst with rust-colored mucoid fluid, which had displaced the floor of the maxillary sinus superiorly (Fig. 9).

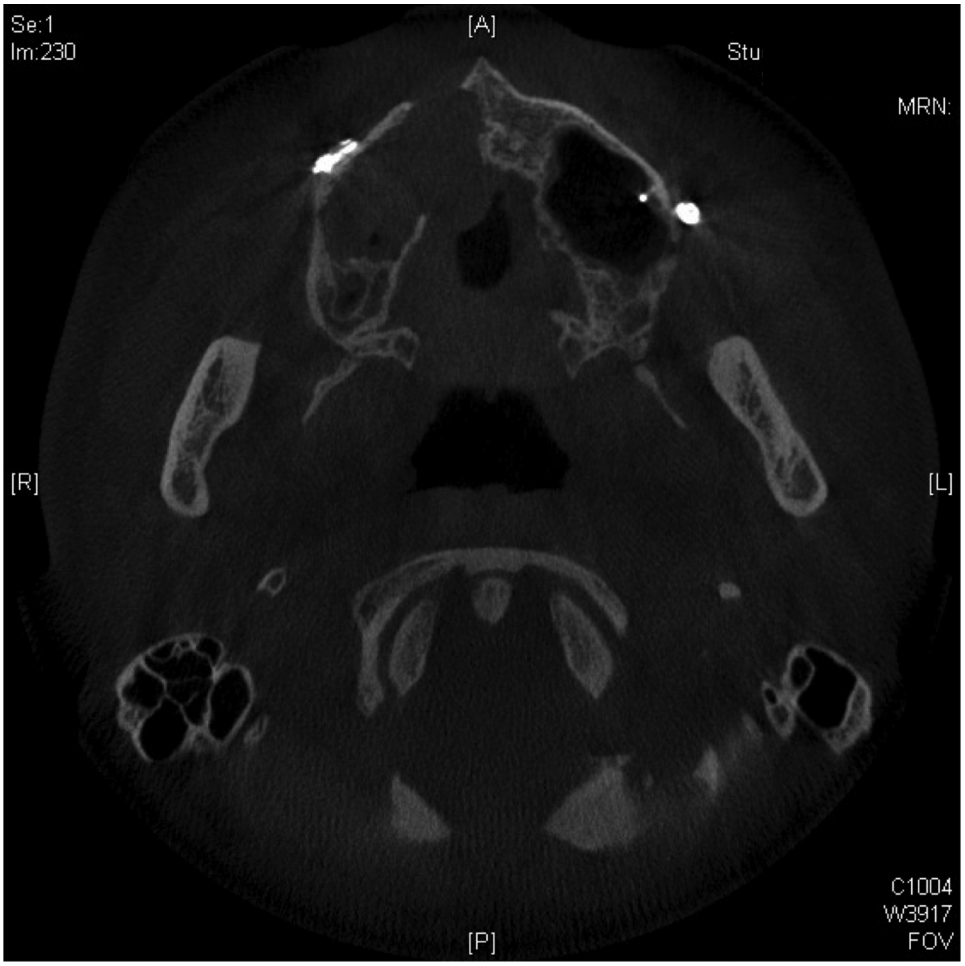

Fig. 6. Axial cone-beam computed tomography showing partial bone defect of the labial and palatal side of the maxilla.

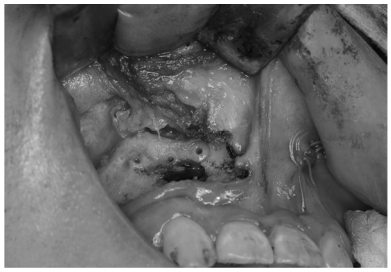

Fig. 8. Thinning of the bone with a perforation in the region of tooth number 13.

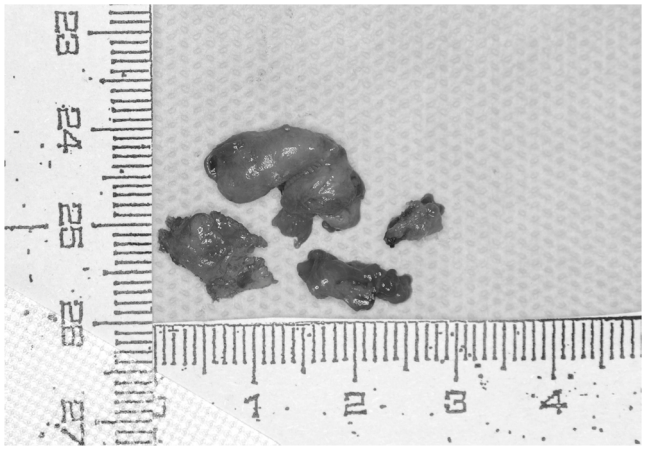

Fig. 9. Excised lesion with smooth wall and rust-colored mucoid content fluid.

The cyst had also expanded and perforated the palatal bone of the maxilla, but the perforation did not appear to be related to any tooth roots. The cyst cavity was subsequently brought into continuity with the residual superiorly displaced maxillary sinus by removing the thin bone that separated the two. Because the nasal side of the bone was resorbed by the cyst, nasal antrostomy was not performed.

Histopathological examination showed a cyst composed of cellular fibrous connective tissue that is mainly lined by stratified squamous epithelium but with regions of focal pseudostratified columnar and ciliated epithelium (Fig. 10). Cholesterin crystal was confirmed within the cyst wall and lymphocyte infiltrate. The diagnosis of surgical ciliated cyst was made on the basis of the histopathological, clinical, and radiographical findings.

Postoperatively, the patient made an uneventful recovery with no sign of recurrence in the following year.

3. Discussion

Surgical ciliated (or postoperative maxillary) cyst is one of the most frequently encountered bony cysts in Japan, with incidences up to 20% of all cysts involving jaws [2–4]. To the best of our knowledge, the occurrence of a surgical ciliated cyst following maxillary orthognathic surgery, such as that reported in the present case, is rare [5,8–14] (Table. 1). Most surgical ciliated cysts occur in the posterior maxilla, typically presenting as an expansive swelling in the maxillary sulcus. Pain and purulent discharge are noted if the region is secondarily infected [2,15]. Radiographs revealed a well-defined, unilocular, radiolucent image with a sclerotic border. It is hypothesized that the pathogenesis of a surgical ciliated cyst is caused by either the entrapment of remnants of sinus mucosa in the wound following a maxillary sinus surgery or an early closure of the natural ostium prior to the sinus being entirely filled with regenerating granulation tissue [2]. The case report by Hayhurst et al[8] postulated that small segments of the nasal mucosa may be entrapped between the bony edges of maxillary osteotomies, resulting in cystic degeneration of the entrapped nasal mucosa many years later. Thus, they recommended that any ruptures in the nasal mucosa should be meticulously repaired, particularly when segmental maxillary surgery is performed.

Surgical ciliated cysts are usually lined by pseudostratified ciliated columnar epithelium of the respiratory type, but transition of such lining to simple columnar, cuboidal, or even squamous epithelium has been observed [2–4]. In the series reported by Sugar et al [5], at the first presentation, one of the cysts had a lining identical to that of antral mucosa. The histological study of 360 cases of surgical ciliated cyst revealed that 66%, 28%, and 6% of the cysts exhibited the lining of pseudostratified columnar ciliated epithelium, transition epithelium, and squamous epithelium, respectively [16]. In the present case, the cyst was mainly lined by stratified squamous epithelium but with regions of focal pseudostratified columnar and ciliated epithelium. The underlying connective tissue was occasionally hyalinized [17].

Yoshiwaka et al [3] discussed the management of surgical ciliated cysts using four different methods: Caldwell-Luc, enucleation with primary closure, enucleation with open packing, and marsupialization. In the 110 cases reported, there were 7 recurrences, and they proposed marsupialization for thin-walled, unilocular cysts with extensive bony perforations. In most cases, enucleation though an approach appropriate to the site will be the treatment of choice. In our case, the cyst had not only expanded but also perforated the palatal bone of the maxilla; however, it did not appear to be related to any tooth roots. The cyst cavity was subsequently brought into continuity with the residual superiorly displaced maxillary sinus by removing the thin bone that separated the two. Nasal antrostomy was not performed because the nasal side of the bone was resorbed by the cyst.

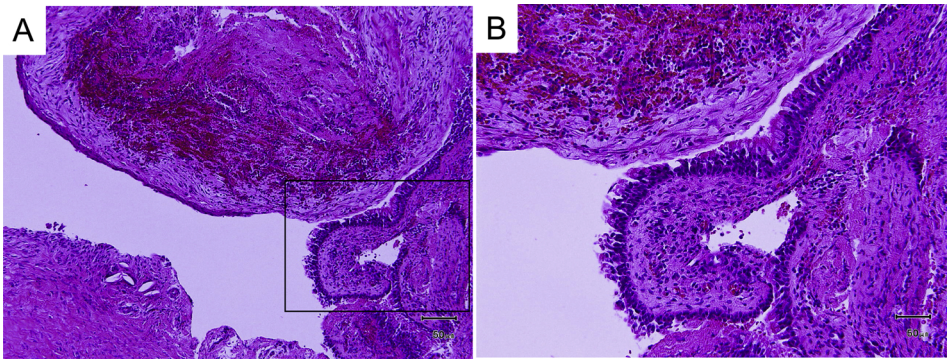

Fig. 10. Histopathology of the surgical ciliated cyst (H & E). (A) Cyst was composed of cellular fibrous connective tissue that is mainly lined by stratified squamous epithelium (100×). (B) There were regions of focal pseudostratified columnar and ciliated epithelium (200×).

Table 1
Previous and present cses of a surgical ciliated cyst occurring following maxillary orthognathic surgery.

	Age	Sex	Surgical History	Fixation	Discovery Period After Surgery	Location	Histopathology	Authors
1	39	F	Le Fort III osteotomy, iliac crest bone grafting	NR	4yr	infraorbital rim	pseuostratified columnar epithelium of respiratory type	Sugar et al [5]
2	21	M	Le Fort II osteotomy, iliac crest bone grafting	NR	3yr	orbital rim	pseudostratified columnar epithelium	Sugar et al [5]
3	38	M	Le Fort I osteotomy, iliac crest bone grafting	NR	3yr	maxillary antrum	pseudostratified columnar epithelium	Sugar et al [5]
4	15	M	Le Fort I osteotomy (midline palatal osteotomy)	NR	5yr	midline of hard palate	ciliated pseudostratified columnar respiratory epithelium	Hayhurst et al [8]
5	17	M	Le Fort I osteotomy	wire fixation	5yr	maxillary alveolus	stratified squamous epithelium, ciliated pseudostratified columnar respiratory epithelium	Amin et al [9]
6	16	F	Le Fort I osteotomy	wire fixation	16yr	maxillary antrum	NR	Thio et al [10]
7	19	F	Le Fort I osteotomy, genioplasty	plate fixation	8yr	mandible	pseudostratified columnar and ciliated epithelium	Bourgeois et al [11]
8	20	M	Le Fort I osteotomy	plate fixation, wire fixation	12yr	maxilla	stratified ciliated columnar or cuboidal epithelium	Takane et al [12]
10	21	M	Le Fort I osteotomy, genioplasty	plate fixation	3yr	chin region	pseudostratified columnar respiratory epithelium	Cai et al [13]
11	18	M	anterior segmental maxillary osteotomy	plate fixation	23yr	maxilla	pseudostratified ciliated epithelium	Kurihara et al [14]
12	25	M	Le Fort I osteotomy	plate fixation	4yr	maxilla	stratified squamous epithelium, pseudostratified columnar epithelium	Present case

NR: not reported.

Although surgical ciliated cysts after orthognathic surgery have scarcely been reported, awareness of such possibility must be spread to avoid delays in diagnosis. Therefore, this lesion should be considered in the differential diagnosis of symptomatic patients who have undergone antral or maxillary orthognathic surgery in the past.

Declaration of Competing Interest

None.

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