Reduction surgery using a combination of a stereolithographic model and navigation system for ossifying fibroma with secondary central giant cell granuloma

Yasuhiro Arai a,*, Yoshihiro Chiba a, Shigeaki Umeda b, Yoshihito Ohara c, Toshinori Iwai c, Masanori Komatsu a, Kenichiro Yabuki a, Daisuke Sano a, Nobuhiko Oridate a

a Department of Otorhinolaryngology and Head and Neck Surgery, Yokohama City University School of Medicine, Yokohama, Japan
b Department of Pathology, Yokohama City University School of Medicine, Yokohama, Japan
c Department of Oral and Maxillofacial Surgery, Yokohama City University School of Medicine, Yokohama, Japan

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A B S T R A C T
Both central giant cell granuloma (CGCG) and ossifying fibroma (OF) are relatively common diseases. The synchronous presentation of CGCG and OF is, however, an extremely rare occurrence. We present an unusual case with the synchronous presentation of these two diseases in the maxilla and introduce a surgical strategy based on a combination of the stereolithographic model and navigation system for the treatment of gigantic OF with secondary CGCG.

Ossifying fibroma (OF) is a well-demarcated benign neoplasm primarily found in the mandible and composed of fibro-cellular tissue and mineralized material showing a different morphologic appearance. OF most commonly occurs in the 2nd to 4th decades and predominantly affects females [7]. OF usually presents clinically as a painless and expansive spherical or ovoid jawbone mass that may displace the roots of adjacent teeth and cause root resorption [8]. Treatment involves the complete removal, as possible, of this usually well-circumscribed tumor, although care should be taken not to sacrifice important adjacent structures and attention given to cosmetic considerations. Tumor recurrence is not uncommon, but growth potential tends to stabilize with time [9]. The synchronous presentation of CGCG and OF is an extremely rare occurrence, although CGCG and OF are both relatively common diseases. Kaplan et al. reported that the CGCG component in combined lesions may shift the clinical behavior toward a more aggressive behavior than that of classical OF [10]. The aim of this report was to present an unusual case showing the synchronous presentation of CGCG and OF in the maxilla and to introduce a surgical strategy based on a combination of a stereolithographic model and navigation system.

1. Introduction
Central giant-cell granuloma (CGCG) is a localized benign, although sometimes aggressive, osteolytic proliferation consisting of fibrous tissue with hemorrhaging and hemosiderin deposits, the presence of osteoclast-like giant cells and reactive bone formation. Most cases are diagnosed in patients less than 30 years of age, with the mandible more often involved than the maxilla. The incidence rate is 1.1 per million per year. In most cases, patients are asymptomatic and diagnosis is made on the basis of incidental findings. Some cases, however, present with pain or paresthesia, swelling, or loosening of teeth [1,2]. It is important that every effort be made to distinguish giant-cell tumors from skull CGCG as giant-cell tumors have a much worse prognosis. Unlike true giant-cell tumors, CGCG has a generally good prognosis with a low incidence of recurrence (12–16%) [3]. The pathogenesis of CGCG remains controversial, with a local reparative process in response to trauma [4], infection [5] and developmental [6] etiology all suggested.

2. Case report
A 44-year-old woman was referred to the Department of Otolaryngology, Head and Neck Surgery, Yokohama City University...
Hospital, due to a painless swelling of the left cheek region of 4 years duration. The patient had experienced acute aortic dissection a month earlier. Her face showed severe asymmetry around the left maxilla area at presentation (Fig. 1A). Oral examination revealed a smooth-surfaced swelling in the left hard palate and periodontal disease (Fig. 1C). High-resolution computed tomographic examination (slice thickness, 0.5 mm) revealed the mass to be 76 mm mediolaterally, 55 mm craniocaudally, and 68 mm anteroposteriorly. It had a central low-density area surrounded by a ground-glass lesion (Fig. 2A). PET-CT scans showed that the ground-glass region surrounding the central low-density area had increased [18F]-fluorodeoxyglucose uptake (maximum SUV, 5.5) (Fig. 2B). As the aortic dissection had become stable after three months of conservative therapy, incisional biopsy through a canine fossa approach was performed under general anesthesia in order to identify its malignant potential. The surface of the tumor was covered with a bony plate. The inner parts bled easily and consisted of a mixture of osseous-fibrous and fragile granulous lesions. Giant-cell granuloma and fibrous dysplasia were suspected on the basis of pathological examinations, and the absence of malignant disease was confirmed. Two months after the incisional biopsy, reduction of the maxilla lesion with preservation of the orbital floor was planned with the aid of a stereolithographic model. DICOM data obtained from computed tomography scanning were processed using Mimics software version 16.1 and 3-matic (Materialise, Belgium). The three-dimensional medical models

![Fig. 1. Facial appearance and oral examination. (A) Facial appearance at the initial presentation. Note the diffuse swelling of the left cheek. (B) A good esthetic outcome was obtained 6 months after the reduction surgery. (C) A smooth-surfaced swelling in the left hard palate and periodontal disease.](image1)

![Fig. 2. (A) CT axial image. CT scans showing the mass to be 76 mm mediolaterally, 55 mm craniocaudally, and 68 mm anteroposteriorly. A central low-density area can be seen to be surrounded by a ground glass lesion. (B) PET-CT image. PET-CT scans showing that the ground glass region surrounding the central low-density area had increased 18F-fluorodeoxyglucose uptake (maximum SUV, 5.5). (C) Simulated views using a canine fossa approach. (D) Simulated views using the Weber–Ferguson approach. (E) T2-weighted MR images showing the central granulation tissue with a well-defined margin.](image2)
were fabricated through a process called powder depositional modeling by use of a ZPrinter 450 3D Color Printer (3D Systems, USA) [11]. Based on this model, we decided that a modified Weber– Ferguson incision, rather than a canine fossa approach, would be suitable for this surgical procedure, which was to be performed using a navigation system (ENT Unlimited 1, Brainlab AG). We were able to obtain a good view of the superior and lateral margins of the tumor by this approach. An electric drill and curette were used to reduce the mass under direct observation of the orbital floor. The central granulation tissue had a well-defined margin on MRI examinations (Fig. 2E). The navigation system allowed us to remove the central granulation tissue while leaving the orbital floor intact.

The operation time was 3 h and 52 min and the total blood loss was 871 ml. Histological examinations showed a circumscribed fibro-osseous lesion consisting of metaplastic bone formation and fibrous stromal tissue. The cellularity was moderate (Fig. 3A). Focal areas were observed with haemosiderin deposits and clusters of multinucleated giant cells (Fig. 3B). These features are consistent with a central giant-cell granuloma arising within a pre-existing ossifying fibroma. Kaplan et al. reported that areas with CGCG characteristics were separated from areas with COF characteristics by a transition zone composed of densely packed spindle cells with varying degrees of collagenization and indistinct vascularity [10]. In the current case, a partial transition zone between the CGCG and OF components was observed (Fig. 3C). However, most of the CGCG and OF components formed a complex contiguous border without any transition zone (Fig. 3D). No remarkable postoperative complications were observed, and the patient was satisfied with the result of this surgery in terms of esthetics (Fig. 1B). At 12 months post-surgery, there was no evidence of tumor enlargement. The periodontal disease was also followed by a dentist. Endoscopic examination at 5 months after surgery showed that the left maxillary sinus was covered with scar tissue (Fig. 4C), and computed tomographic examination at 6 months after surgery revealed subtotal resection of the central granulation tissue (Fig. 4A, B). Regular follow-up of the patient was indicated.

3. Discussion

Seven cases of central giant-cell granuloma (CGCG) associated with ossifying fibroma (OF) have been reported [10,12–15], with the mass located in the mandible and maxilla [10,14] in 5 and 2 of them, respectively. Three of the 7 cases were controlled with conservative therapy (reduction surgery [10,13] or calcitonin spray [10]).

With regard to the etiology, Kaplan et al. reported that the primary lesion could be an ossifying fibroma in which, through some as yet unknown trigger, the mesenchymal spindle cells of the tumor release cytokines that induce differentiation toward osteoclast/giant cells. This results in the formation of the CGCG component [10]. In the current case, a local reparative process in response to infection associated with periodontal disease might have been the trigger. The mass observed in this case report was much larger than those previously reported. Therefore, we first conducted an open biopsy to identify its malignant potential. Following confirmation of the absence of malignancy, we selected reduction surgery to minimize the patient’s burden. Simulation using a stereolithographic model aided in the performance of safe and timely surgery in that this model allowed us to preoperatively view the craniofacial image in three dimensions from various angles. During simulation, we realized that we could not obtain a good view of the superior and lateral margins of the tumor by a canine fossa approach (Fig. 2C). We also realized that there was a

![Fig. 3](image-url) (A) A circumscribed fibro-osseous lesion consisting of metaplastic bone formation (black arrow) and fibrous stromal tissue (black arrowhead). Original magnification is ×100. (B) Focal areas with haemosiderin deposition and clusters of multinucleated giant cells (black arrow). Original magnification is ×100. (C) A partial transition zone between CGCG and OF components was observed. Original magnification is ×40. (D) Most of the CGCG and OF components formed a complicated contiguous boundary without any transition zone. Original magnification is ×40. OF: ossifying fibroma, CGCG: central giant-cell granuloma, T: transition zone.
risk of the orbital floor collapsing under a Weber–Fergusson incision (Fig. 2D) as we found the lesion extended to the anteroinferior orbital wall and the inferior rim of the orbital wall could be fragile at the previous incisional biopsy. We, therefore, added an incision along lower eyelid to the Weber–Fergusson incision and were able to outline the entire tumor. The other reason for this was to avoid massive bleeding as the previous incisional biopsy had revealed that the tumor bled easily. We judged that the surgery would be more time-consuming and could cause massive bleeding under a limited surgical view and working space such as that afforded through a canine fossa approach. In general, the subtotal resection of the tumor through a canine fossa approach may not be difficult where a lengthy surgical procedure and unexpected blood loss are acceptable. However, the patient had had an aortic dissection five months prior to the reduction surgery and the cardiologist advised us that the circulatory dynamics needed to be stabilized during the perioperative period. Under such conditions, a lengthy surgical procedure and unexpected blood loss had to be avoided. We discussed this complication with the cardiologists and anesthesiologists and concluded that a modified Weber–Fergusson approach would be the best approach for this patient in order to decrease the surgical time and blood loss. We then performed this reduction surgery under direct observation of the orbital floor with an imaginary original bony configuration. A combination of a stereolithographic model and navigation system helped us to save operative time as well as avoid heavy bleeding. No remarkable postoperative complications were observed.

As the CGCG component in the lesions may lead to a more aggressive clinical behavior than classical OF [10,14], the minimum goal of the reduction surgery was the complete removal of the CGCG component. Use of the navigation system allowed us to successfully remove the central granulation tissue while leaving the orbital floor intact. However, Kaplan et al. reported that it is impossible to determine whether different lesions containing CGCG-like areas, including combined lesions, share the potential for the aggressive biologic behavior observed in cases of CGCG without further studies. Furthermore, one [10] of the 3 previously reported cases treated by reduction surgery [10,13] experienced recurrence within 12 months. Therefore, it is recommended that cases of combined tumors be followed closely [10].

4. Conclusion

Surgical reduction using a combination of a stereolithographic model and navigation system was effective in the treatment of this gigantic case of ossifying fibroma with secondary central giant-cell granuloma in terms of both surgical time and bleeding.

Conflict of interest

We have no conflicts of interest to declare with regard to this manuscript.

References


