Modified Lothrop (Draf III) procedure for the treatment of a recurrent orbitofrontal cholesterol granuloma: A case report

Hidenori Yokoi1, Hidetaka Yamanaka1, Yuma Matsumoto1, Michitsugu Kawada1, Masachika Fujiwara2, Arisa Ohara3 and Koichiro Saito1

Abstract
Orbitofrontal cholesterol granuloma is a rare occurrence. Here, we present a case involving a 64-year-old man with a recurrent orbitofrontal cholesterol granuloma treated by the Modified Lothrop (Draf III) procedure. The patient, who had a history of trauma and previous sinus surgery, presented with chief complaints of nasal congestion, olfactory impairment, and diplopia. We suspected chronic sinusitis; computed tomography showed a soft-tissue shadow extending from the bilateral frontal sinuses to the ethmoid sinuses, with a cyst in the right orbitofrontal region. We performed endoscopic surgery for removal of the mass, and histopathological analysis of the resected specimen confirmed a diagnosis of cholesterol granuloma. The lesion recurred 2 months later, and we performed revision surgery using the Modified Lothrop or Draf III procedure. The patient showed no relapse at the 5-year follow-up. These findings suggest that the Draf III procedure is an effective surgical treatment for cholesterol granulomas.

Keywords
Sinusitis, cholesterol granuloma, orbitofrontal, trauma, Draf III procedure

Date received: 20 December 2018; accepted: 21 January 2020

Introduction
Cholesterol granuloma is a histopathological entity thought to be caused by a chronic granulomatosis reaction to the presence of cholesterol crystals released by the breakdown of blood and local tissue in enclosed spaces.1,2 The lesion is histopathologically characterized by foamy histiocytes, giant cells, plasma cells, lymphocytes surrounding cholesterol clefts, and hemosiderin deposition.3 Obstructed drainage and impaired ventilation are considered significant etiological factors.4 Cholesterol granulomas most commonly develop in the mastoid portion of the temporal bone and middle ear.5 Here, we report a rare case involving an elderly man with a cholesterol granuloma in the right orbitofrontal region that possibly developed after facial trauma. The granuloma was initially removed via endoscopic endonasal surgery (ESS), but it recurred and was successfully treated using the modified Lothrop (Draf III) procedure.

Case report
A 64-year-old man presented with chief complaints of nasal congestion, olfactory impairment, and diplopia. At 31 years of age, he had undergone the Caldwell–Luc procedure for chronic paranasal sinusitis. At 56 years of age, a bicycle accident resulted in right frontal sinus contusion with right eyeball protrusion, which was left untreated. Approximately 8 years later, he experienced diplopia during upward gazing and was referred to our hospital.

On presentation, the patient’s visual acuity was normal, but the right eye was positioned lower than the left one, and diplopia was present. Computed tomography (CT) revealed a soft-tissue shadow extending from the bilateral frontal sinuses to the ethmoid sinuses. We also observed a cyst progressing...

1Department of Otolaryngology, Head and Neck Surgery, Kyorin University School of Medicine, Tokyo, Japan
2Department of Pathology, Kyorin University School of Medicine, Tokyo, Japan
3Department of Radiology, Kyorin University School of Medicine, Tokyo, Japan

Corresponding Author:
Hidenori Yokoi, Department of Otolaryngology, Head and Neck Surgery, Kyorin University School of Medicine, 6-20-2 Shinkawa, Mitaka 181-8611, Tokyo, Japan.
Email: h-yokoi@ks.kyorin-u.ac.jp
toward the right orbitofrontal region, with thinning of the wall of the right ethmoid orbital plate (Figure 1).

Magnetic resonance (MR) imaging showed a mass at the top of the right orbital cavity, which passed through the inner wall and protruded into the orbital cavity. The mass showed a high-intensity signal on both T1- and T2-weighted images, with a slightly less intense signal on the T2-weighted images. As a result, the superior rectus muscle is compressed and displaced to the right, and there is protrusion of the right eyeball. Accumulation of low-signal intensity contents thought to be mucosal thickening and concentrated contents can be observed in the frontal and ethmoid sinuses. (c) Contrast-enhanced T1-weighted image. A well-defined space-occupying lesion with smooth margins is found advancing into the right orbit from the right frontal and ethmoid sinuses. The T1-weighted image before contrast enhancement already exhibits a high signal intensity, and contrast enhancement does not result in clear internal intensification on a subtraction image.

This suggests that the lesion is primarily a liquid cystic lesion. There is no inferolateral displacement of the right orbital contents, and no clear abnormal enhancement of the adjacent mucosal thickening or cerebral parenchyma (right frontal lobe).

Considering the imaging findings and a diagnosis of recurrent sinusitis, we performed ESS involving bilateral ethmoidectomy. Following removal of the chronically inflamed mucous membrane, the nasofrontal ducts were easily identified and the bilateral frontal sinuses were thoroughly drained. Histopathological analysis of the orbitofrontal lesion revealed characteristic separation of the epithelium, inflammatory cell infiltration in the stroma, hyalinized collagen fibers in the cyst wall, hemosiderin deposits, and
cholesterol crystals, features indicative of cholesterol granuloma (Figure 3). Another pathological mucosal sample showed nonspecific inflammation.

The patient’s eye position normalized and diplopia resolved after surgery. However, follow-up CT after 2 months showed recurrence of a soft, dense mass in the same area. Therefore, we decided to perform revision surgery using the Draf III procedure. More specifically, we performed frontal sinusotomy, enlarged a perforation in the nasal septum adjacent to the frontal sinuses, and widely opened the frontal sinuses to create a single lumen into the bilateral ethmoidal sinuses (Figure 4(a)). Follow-up CT after 5 years showed no recurrence of the cholesterol granuloma (Figure 4(b)).

The patient provided written consent for data publication. Ethics committee approval was waived.
Discussion

It has been suggested that cholesterol granulomas develop when chronic inflammation occurs in a pneumatic space and blocks the air circulation. When there is additional surgery or bleeding, blood or exudate accumulates within the blocked cavity, leading to the separation and precipitation of cholesterol in the form of crystals composed of protein–lipid conjugates. Inflammatory cells further accumulate in the area as part of the foreign body reaction to these crystals, and a granulomatous structure is formed.\(^1,6\)

The most probable cause for cholesterol granuloma formation within a closed cavity is a history of surgery or trauma. However, morphological paranasal sinus abnormalities can also result in the formation of granulomas, particularly in the orbital cavity or frontal sinuses.\(^7\) Our patient had abnormal paranasal sinus morphology due to previous trauma, surgery for chronic sinusitis, and recurrence of sinusitis. He exhibited an ocular motility disorder, eyeball protrusion, diplopia, and clinical and radiological findings consistent with those previously reported for orbitofrontal cholesterol granuloma.\(^4\) Taken together, our findings suggest that the formation of cholesterol crystals in this patient was due to the hemorrhagic transformation in the residual mucosa following the trauma or sinus surgery.

CT imaging of cholesterol granulomas often reveals a soft-tissue shadow and bone breakdown. These lesions reportedly appear as high-intensity signals with no contrast effect on T1- and T2-weighted images.\(^5\) In our case, CT findings suggested that the bilateral maxillary sinuses had narrowed because of the Caldwell–Luc procedure performed for chronic sinusitis 33 years back. We also observed a soft-tissue shadow in the ethmoid and frontal sinuses, recurrent sinusitis, and a right orbitofrontal cyst. Moreover, T1- and T2-weighted images showed a variety of intensities at the lesion site, possibly due to differences in the protein concentration. In retrospect, we should have strongly suspected an orbitofrontal cholesterol granuloma on the basis of these MR imaging findings.

Common treatments for orbitofrontal cholesterol granulomas include drainage with perforation and surgical incisions for smaller and larger lesions, respectively. A number of reports have documented the use of craniotomy or orbitotomy through the glabella\(^10\) and, more recently, endoscopic procedures.\(^2,11\) During conventional endoscopic frontal surgery, the extranasal frontal sinus path is selected in cases where the drainage pathway cannot be sufficiently enlarged, which results in restenosis.\(^2,11\) However, extranasal surgery is highly invasive and increases the likelihood of postoperative cyst formation when there is insufficient air exchange within the nasal cavity. Previous studies showed favorable results after frontal sinus opening using the Draf III procedure. Since then, this method has become a standard approach for refractory frontal sinusitis management and is less invasive than the extranasal approach.\(^12,13\)

Minimally invasive treatments for cholesterol granulomas reportedly lower the recurrence rate.\(^14\) For our patient, we initially selected ESS to widely open the nasal–frontal sinus tract on the basis of the radiological findings and recurrent sinusitis diagnosis. However, restenosis occurred after 2 months. Because the excised orbitofrontal lesion had been diagnosed as a cholesterol granuloma, we performed revision surgery using the Draf III procedure. Because cholesterol granulomas rarely involve the mucosal epithelium of the paranasal sinuses, which contain air, their recurrence rate after cyst formation or in closed cavities is low, provided there is no postoperative residual mucosal epithelium. A previous follow-up study of patients with orbitofrontal cholesterol granulomas reported recurrence in seven of 97 patients, which was presumably caused by orbital surgical incisions in all patients. However, there was no mention of the efficacy of the endoscopic surgery. Recently, however, Curtis et al.\(^15\) reported that Draf II procedure were effective for frontal sinus cholesterol granuloma. In our case, we managed to create an opening at the nasal septum, at the site adjoining the frontal sinuses, through frontal sinusotomy. This resulted in the formation of a large open space, which enabled complete removal of the pathological mucosa. Accordingly, we believe that the Draf III procedure is quite effective for treating orbitofrontal cholesterol granulomas.

Conclusion

To our knowledge, this is the second case of orbitofrontal cholesterol granuloma successfully treated using the Draf III procedure in the English literature. Nevertheless, we consider this approach effective for the surgical treatment of cholesterol granulomas, even when the pathology is not limited to the orbitofrontal region and recurrent ethmoid sinusitis does not present a risk. The Draf III procedure permits the efficient opening of the blocked cavities, thus allowing for a wider field of view and complete removal of the affected mucosa. In summary, our findings suggest that the Draf III procedure can be used to complement conventional ESS for the successful surgical treatment of orbitofrontal cholesterol granulomas.

Acknowledgements

We would like to thank Editage (www.editage.jp) for English language editing.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.
Funding
The author(s) received no financial support for the research, authorship, and/or publication of this article.

Informed consent
Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

ORCID iD
Hidenori Yokoi https://orcid.org/0000-0001-7058-3553

References
1. Niho M. Cholesterol crystals in the temporal bone and the paranasal sinuses. Int J Pediatr Otorhinolaryngol 1986; 11(1): 79–95.
2. Weiland DA and Aygun N. An unusual presentation of a cholesterol granuloma in a pneumatized pterygoid process of the sphenoid sinus. Otolaryngol Head Neck Surg 2007; 136(1): 153–154.
3. Hwang DJ, Chung YS, Jun SY, et al. A case of compressive optic neuropathy caused by sphenoid sinus cholesterol granuloma. Jpn J Ophthalmol 2009; 53(4): 441–442.
4. Almada CB, Fonseca DR, Vanzillotta RR, et al. Cholesterol granuloma of the maxillary sinus. Braz Dent J 2008; 19: 171–174.
5. Karaky AA, Sawair FA, Baqain ZH, et al. Cholesterol granuloma of the maxillary sinus encountered during floor augmentation procedure: a case report. Clin Implant Dent Relat Res 2010; 12(3): 249–253.
6. Shykhn ME, Trotter MJ, Morgan DW, et al. Cholesterol granuloma of the frontal sinus. J Laryngol Otol 2002; 116: 1041–1043.
7. Hill CA and Moseley IF. Imaging of orbitofrontal cholesterol granuloma. Clin Radiol 1992; 46(4): 237–242.
8. Hughes JD, Jacob JT, Garrity JA, et al. Orbitofrontal cholesterol granuloma: four case reports and a systematic review of the English literature. World Neurosurg 2016; 87: 355–361.
9. Morioka T, Fujii K, Nishio S, et al. Cholesterol granuloma in the middle cranial fossa: report of two cases. Neuroradiology 1995; 37(7): 564–567.
10. Selva D and Chen C. Endoscopic approach to orbitofrontal cholesterol granuloma. Orbit 2004; 23(1): 49–52.
11. Sia DI, Davis G and Selva D. Recurrent orbitofrontal cholesterol granuloma: a case report. Orbit 2012; 31: 184–186.
12. Deep NL, Chaaban MR, Chaudhry AL, et al. Frontal sinus cholesterol granuloma: case report. Allergy Rhinol (Providence) 2014; 5: 36–38.
13. Eloy P, Vlaminck S, Jorissen M, et al. Type III frontal sinusotomy: surgical technique, indications, outcomes, a multi-university retrospective study of 120 cases. B-ENT 2011; 7(Suppl. 17): 3–13.
14. Durgam A and Batra PS. Paranasal sinus cholesterol granuloma: systematic review of diagnostic and management aspects. Int Forum Allergy Rhinol 2013; 3(3): 242–247.
15. Curtis S, Butrynnowicz A, Kenning TJ, et al. The modified Lothrop procedure (Draf III frontal sinusotomy) in the management of frontal sinus disease: a patient outcomes study. J Neurol Surg B 2016; 77: P038.