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

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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

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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

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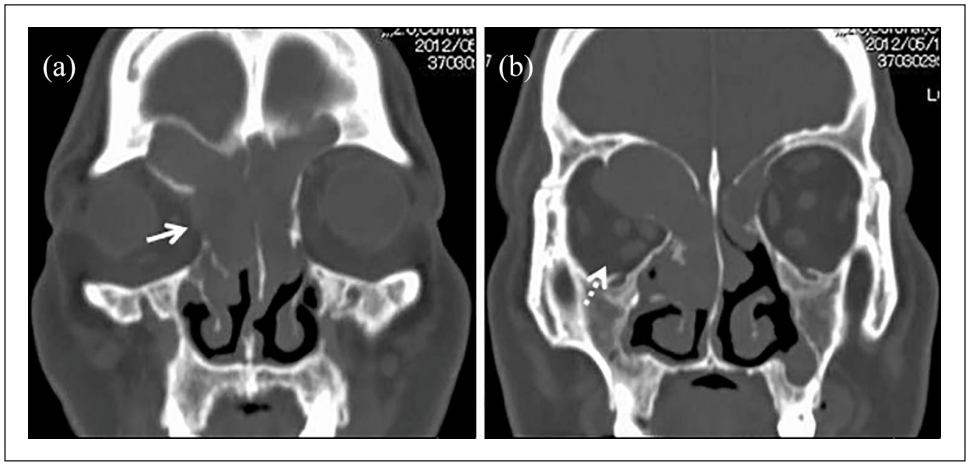

Figure 1. Preoperative computed tomography images for an elderly man with an orbitofrontal cholesterol granuloma. (a) Coronal views show a well-defined space-occupying lesion exhibiting expansion and displacing growth, advancing from the right frontal and ethmoid sinuses to the right orbit and right nasal cavity. There is no clear bone breakdown caused by this lesion, and the bone exhibits marked thinning at one site (white arrow). (b) Because of advancement into the right orbit, the orbital contents are compressed and displaced laterally and inferiorly (white dotted arrow). A soft structure is also observed in the left frontal sinus and left nasal cavity, with marked thinning of the bone at one site. No clear advancement into the left orbit can be seen.

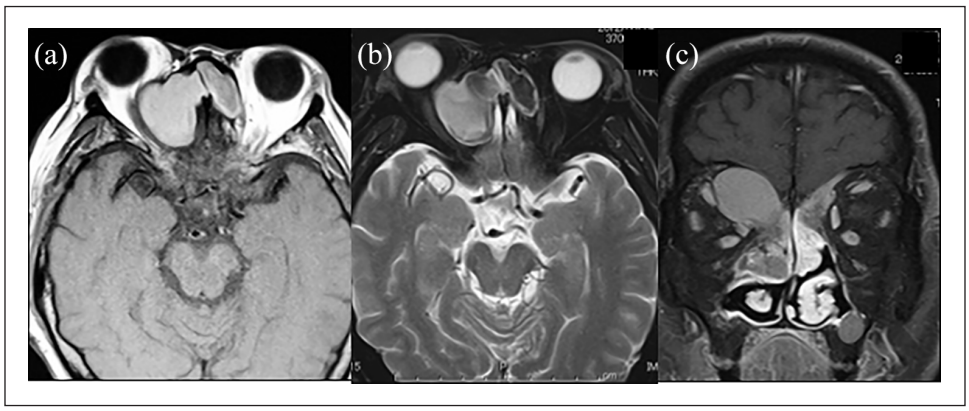

Figure 2. Preoperative magnetic resonance imaging findings for an elderly man with an orbitofrontal cholesterol granuloma: (a) T1-weighted image and (b) T2-weighted image. An elliptical mass with smooth margins can be seen extending from the right ethmoid sinus into the orbit. The mass shows a high-intensity 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).

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. The left middle turbinate was swollen, and polypoid degeneration was suspected (Figure 2).

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

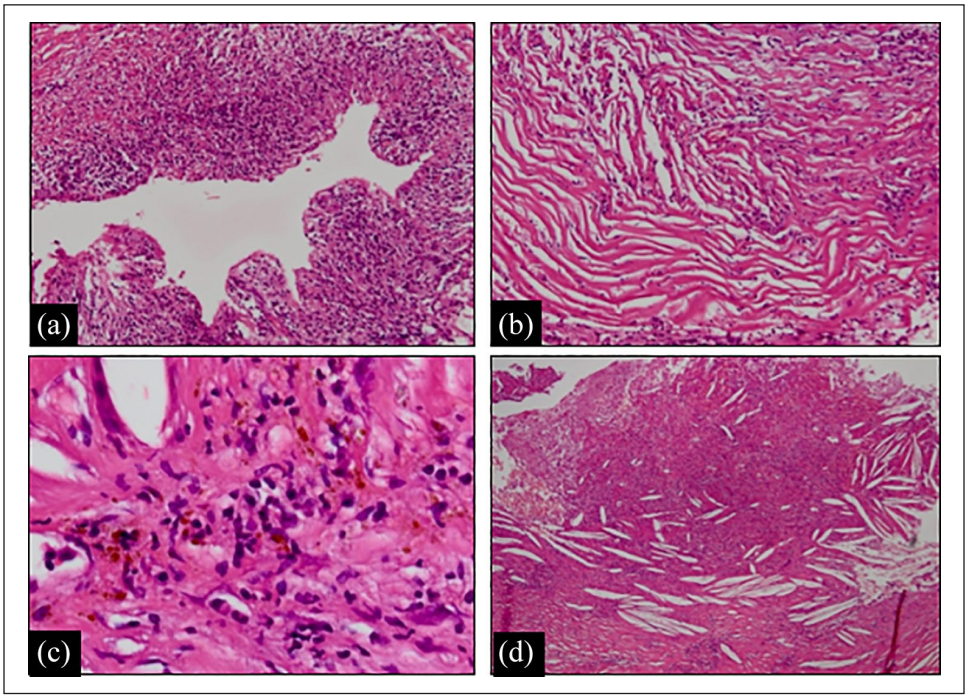

Figure 3. Histopathological analysis of an orbitofrontal cholestrol granuloma endoscopically resected from an elderly man (hematoxylin and eosin staining): (a) separation of the epithelium and infiltration of inflammatory cells in the stroma, (b) hyalinized collagen fibers in the cyst wall, (c) hemosiderin deposits, and (d) cholesterol crystals.

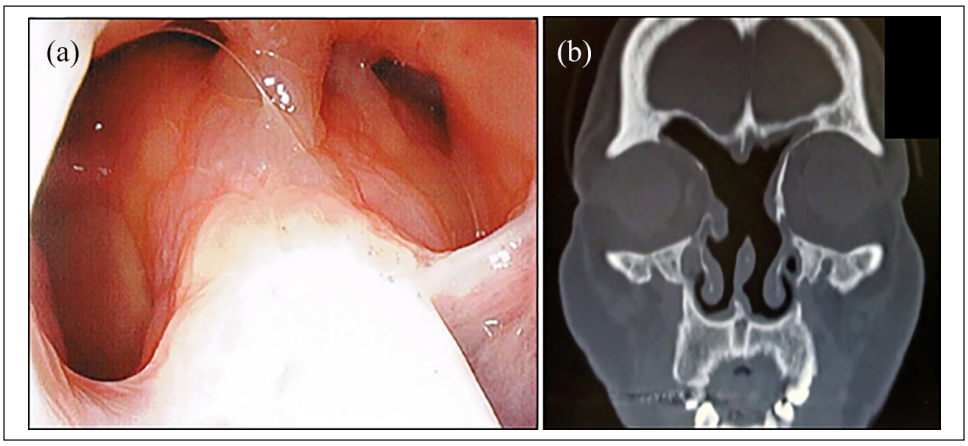

Figure 4. Postoperative view and computed tomography image for an elderly man who underwent the Draf III procedure for a recurrent orbitofrontal cholesterol granuloma: (a) photograph showing the region of the bilateral frontal sinuses after the Draf III procedure, and (b) computed tomography image (coronal view) showing the bilateral ethmoid sinuses and frontal cavity. There is no residual morbid soft tissue.

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.[8] 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.[9] 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.[10,11] 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.[10,11] 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.[8] 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.

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Ethical approval

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Informed consent

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