CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 76 (2020) 263–265

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Xanthogranuloma formation after endoscopic sinus surgery: A case report

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ARTICLE INFO

Article history:
Received 15 September 2020
Received in revised form
29 September 2020
Accepted 29 September 2020
Available online 2 October 2020

Keywords:
Xanthogranuloma
Endoscopic sinus surgery
Blow out fracture

ABSTRACT

INTRODUCTION: Although xanthogranuloma is known to be related to trauma or mucosa, possibly developing around a periorbital or oral lesion, xanthogranuloma related to sinusitis urgery has not been reported. We present a case of xanthogranuloma formation after endoscopic sinus surgery (ESS).
PRESENTAION OF CASE: A 54-year-old man with pain and swelling in the right periorbital area presented to our clinic. He had had a blowout fracture treated by ESS 2 years prior. Physical examination and computed tomography revealed an ~1-cm × 0.7-cm cystic mass on the right lower eyelid. Subciliary exploration found a fat-like mass that we completely excised. A histological examination revealed xanthogranuloma. No recurrence was observed for 1 year.
DISCUSSION: If the wall between the sinuses and the orbit and the mucosa of the maxillary sinus are injured during ESS, infectious material and hematoma could develop into chronic granulomatous inflammation. In addition, a large antrostomy and/or a damaged nasolacrimal duct are risk factors for xanthogranuloma. Antibiotics can treat the disease and prevent infection. Progressive growth of the lesion and its infiltration into surrounding tissues may result in surgical resection.
CONCLUSION: Because many masses are idiopathic, the development of xanthogranuloma after simple ESS or a nondisplaced blowout fracture is possible. Although xanthogranuloma progression usually is benign and without specific complications, it may be sight- or life-threatening. Antibiotics and surgical resection are the treatments of choice and the latter can be a diagnostic tool. Physicians should be aware of the possibility of granuloma formation in patients who have undergone ESS.

1. Introduction

Xanthogranuloma is a rare inflammatory granulomatous lesion that is found mostly in the mediastinum, retroperitoneum, lymph nodes, and bones [1]. Early histopathologic signs of xanthogranulomas include large collections of banal-appearing histiocytes in the dermis, sometimes extending into subcutaneous fat and even the fascia. Xanthogranuloma is associated mainly with trauma, infection, and malignancy, but its causal mechanism is little understood [1–4]. The rate of orbital complications after endoscopic sinus surgery (ESS) is very low, with orbital edema after fracturing the lamina papyracea the most common [5]. Although xanthogranuloma is known to be related to trauma or mucosa and usually develops around a periorbital or oral lesion, there have been no reports of it being related to sinusitis surgery. Our study was reported following SCARE 2018 guidelines [6]. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request and all photograph in this article was permitted by patient having a signed consent form to be publicated. We present a case of xanthogranuloma formation after endoscopic sinus surgery.

2. Presentation of case

A 54-year-old man presented to our clinic complaining of pain and swelling in the right periorbital area. He had undergone endoscopic sinus surgery (ESS) for chronic sinusitis about 2 years prior to presentation. His orbits were severely swollen on postoperative day 2 after blowing his nose. We ordered computed tomography (CT) which showed that the patient had blowout fracture complications after the ESS and emphysema but no defects. The patient had not undergone any additional surgery or treatment. We assumed that the orbital injury developed after the ESS. The swelling recurred over 2 years without any symptoms. However, the patient found a focal mass on his lower eyelid 6 months before revisiting our clinic. He denied any history of drug use, family history, including any relevant genetic information, and psychosocial history. Our physical examination of the patient found a soft, movable, and palpable mass approximately 1 cm in diameter on the right lower eyelid (Fig. 1). Computed tomography revealed a 1-cm × 0.7-cm cystic mass in the right lower eyelid region and demonstrated the status

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https://doi.org/10.1016/j.ijscr.2020.09.200

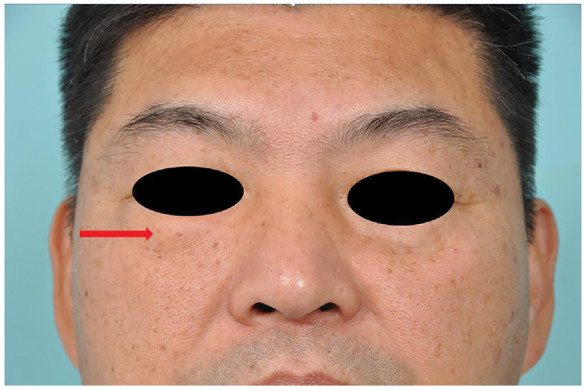

Fig. 1. The mass was palpable in the right lower eyelid, and was movable, non-tender and rounded. (Red Arrow).

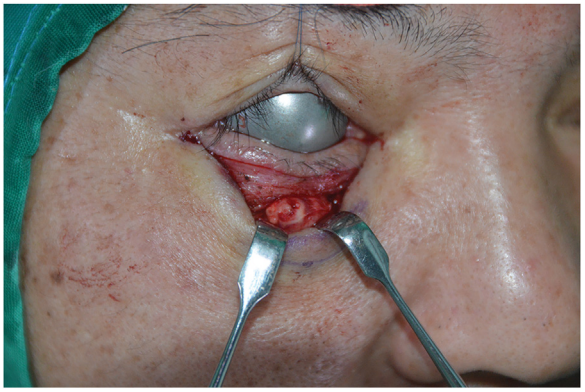

Fig. 3. About 1 cm sized cystic mass was found between orbicularis orbital muscle and orbital septum.

after ESS (Fig. 2). The mass was already localized, so we decided to examine it using a subciliary approach. A plastic surgeon performed the surgery and found a graish tan colored lipomatous mass on the right lower eyelid (Fig. 3). The patient tolerated the surgery and was discharged afterward. The mass was completely excised and sent to the pathology laboratory. The histological examination found that there were some variable-sized fat-containing pseudocystic granulomas (Fig. 4) and the mass was consistent with xanthogranulomatous inflammation. The mass was benign and did not need further treatment. The patient completed 2 weeks of oral antibiotics and no recurrence was observed for 1 year.

3. Discussion

During ESS, it is not always possible to avoid injuring the extremely thin bony wall between the sinuses and the orbit, possibly causing maxillary sinus mucosa or other infectious materials and hematoma to remain. Because these materials cannot be degraded by cellular enzymes, they act as foreign bodies, triggering diffuse chronic granulomatous inflammation. There are several reported cases of mass lesions that occurred after ESS that were related to paraffin-induced sclerosing lipogranuloma. Witschel and Geiger [7] reported on 10 patients who developed paraffin-induced sclerosing lipogranuloma over 5 years, while other authors [8,9] reported lipogranuloma induced by the use of paraffin or Vaseline following nasal packing or ointment use. In addition, Raymond [10] and Malik et al. [11] reported cases of xanthogranuloma in the maxillary sinus; however, it was not related to ESS but to tooth extraction and sinusitis, respectively. In these cases,

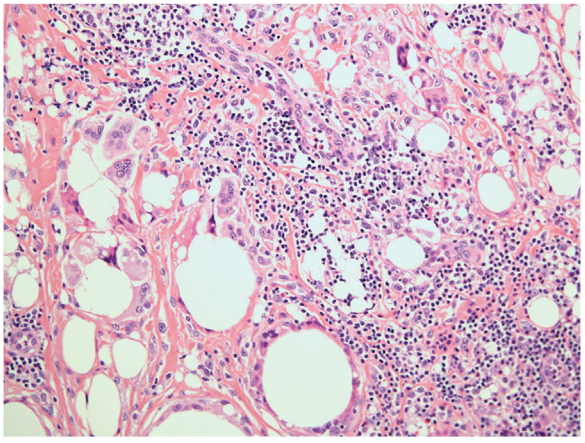

Fig. 4. Pathologic image of the mass located in the lower eyelid are gray tan tissue measuring 1.0 × 0.7 cm (H&E stain, ×200). The lesion is consistent with xanthogranuloma inflammation with variable sized fat containing pseudocystic granulomas.

the ENT surgeon used Merocel (Medtronic Inc., Minneapolis, MN, USA), a compressed, dehydrated sponge composed of hydroxylated polyvinyl acetate and one of the most common nonabsorbable nasal packing materials. Xanthogranuloma is a rare and inflammatory granulomatous lesion, the pathogenesis of which is not well understood thus far. However, most theories on its etiology link it to inflammation. Antibiotics are used to treat the disease and to prevent infections. Progressive growth of the lesion and its infiltration of the surrounding tissues may result in surgical resection as a treat-

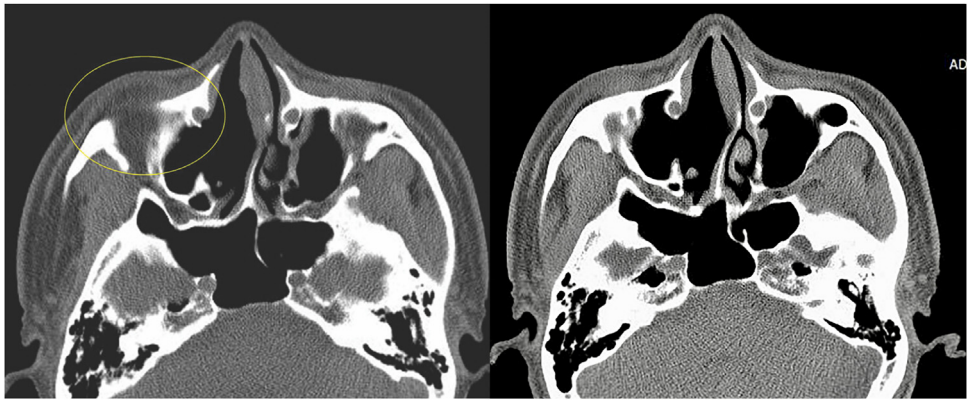

Fig. 2. The computed tomography (CT) demonstrated subcutaneous cystic mass lesion on right lower eyelid lesion measuring about 1 cm (Circle, Left). Definite fracture such as blow out fracture or empyema was not identified at this stage and endoscopic sinus surgery was proved by this image. Post endoscopic sinus surgery CT showed large antrostomy with undamaged nasolacrimal duct (Right).

ment option and diagnostic tool at the same time. If completely removed, the lesion does not recur [11].

We completely resected the lesion and treated the patient with intravenous antibiotics for 3 days, followed by oral antibiotics. The patient healed and there was no recurrence for a year. Because we believed that the mass had already localized, we assumed that complete excision was the best method for diagnosis and treatment. However, medical treatment alone possibly could have resolved this lesion. It was not clear if the lesion was causally related to the ESS. Therefore, we are only suggesting possibilities. This uncertainty is a limitation of this study. Because many masses are idiopathic, it is possible that xanthogranuloma could develop after simple ESS or a nondisplaced blowout fracture. In addition, a large antrostomy and/or a damaged nasolacrimal duct are a risk factors for development of xanthogranuloma. Of the 4 types of xanthogranuloma [12,13], our case seemed to be the adult onset xanthogranuloma type (i.e., solitary lesion without systemic findings). Although this type usually progresses benignly and has no specific complications, it may be sight- or life-threatening. Therefore, its recognition by the ophthalmologist is critical because periocular features are often the initial presentation [13].

Conclusion: Because many masses are idiopathic, we suggest the possibility that xanthogranuloma develops after simple ESS or a nondisplaced blowout fracture, especially if there was a large antrostomy and the nasolacrimal duct was damaged. Although progression of this lesion is usually benign and without specific complications, it may be sight- or life-threatening. Therefore, surgeons should be aware of the possibility of granuloma formation in patients who have undergone ESS for paranasal sinus disease.

Declaration of Competing Interest

The authors have nothing to disclose.

Funding

No funding was received for this article.

Ethical approval

Not applicable. (one case who underwent usual treatment, we got a patient consent and agreement for publication).

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ja Hea Gu: Study concept or design, Data analysis and interpretation, Confirmation of final paper

Gyu Hyeong Lee: Writing the draft, data collection

Registration of research studies

This article is not human studies. Report of one case who underwent usual treatment.

Guarantor

Ja Hea Gu has a full responsibility for the work and the conduct of the study, access to the data and controlled the decision to publish.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgement

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

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