

Bilateral cholesterol granulomas of the maxillary sinus with review of the literature

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Nicole Wershoven^{1,2} , Courtney Miller^{1,2}, Pamela Gibson^{1,3}, Allison Ciolino^{1,3} and Tamara Rimash^{1,2}

Abstract

Cholesterol granulomas are a common benign pathology classically found in the mastoid antrum and air cells of the temporal bone and less commonly found in the paranasal sinuses. We present a unique case of bilateral cholesterol granulomas of the maxillary sinus that is the second case to our knowledge reported in the literature. In an effort to provide an update about cholesterol granulomas of the paranasal sinuses, we examined the literature from January 2011 through 2021 in conjunction with a previous systematic review of the literature from 1970 to December 2010. This report reinforces that upon presentation, cholesterol granulomas can resemble multiple pathologies and histology is needed for diagnosis. This report should serve as an updated resource for otolaryngologists regarding cholesterol granulomas of the paranasal sinuses.

Keywords

Allergy/immunology, otolaryngology, pathology, rhinology, nasal mass, cholesterol granuloma, paranasal sinus

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Introduction

Cholesterol granulomas are a common benign pathology classically found in the mastoid antrum and air cells of the temporal bone and less commonly found in the skull and paranasal sinuses.^{1,2} They are precipitated cholesterol clefts that ensue a foreign body reaction.³ Exact pathogenesis is unknown but it has been hypothesized to be due to impaired drainage, poor ventilation, or hemorrhage in the sinus. There are two predominating theories of formation which include the Obstruction-Vacuum theory and the Exposed Marrow theory. The Obstruction-Vacuum theory, which is the older theory, states that the combination of mucosal swelling and resorption of air causes a negative pressure environment that subsequently causes hemorrhage and ensues an inflammatory reaction.⁴ The Exposed Marrow theory hypothesizes that excessive pneumatization of bone causes marrow to be exposed which can provide a hemorrhagic milieu.⁴ The cholesterol results from the breakdown of the red blood cell membrane and connective tissue breakdown due to poor oxygen supply from obstruction.^{5,6} The crystallized cholesterol stimulates a granulomatous reaction that attracts inflammatory cells such as leukocytes and macrophages.^{5,6} Patients typically present with symptoms of yellowish rhinorrhea, nasal obstruction, facial pain, headache sinusitis, and rhinitis.^{1,7} Due to the mimicking nature and rarity of cholesterol granulomas in the paranasal sinuses, they are

often confused with entities such as mucocoele, neoplasm, odontogenic cyst, aneurysmal bone cysts, or ossifying fibroma.^{1,3} As of 2020, only 51 cases of cholesterol granulomas in the maxillary sinus have been reported in literature. This is the second case to our knowledge of bilateral maxillary sinus cholesterol granulomas. This report and literature review should serve as an updated resource for otolaryngologists regarding cholesterol granulomas of the paranasal sinuses and an imaging reference for bilateral lesions.

Case report

A 50-year-old female presented to clinic for evaluation of sinusitis with nasal polyposis. Upon evaluation, she noted symptoms of facial pressure, dental pain, nasal congestion,

¹The Robert Larner, M.D. College of Medicine, The University of Vermont, Burlington, VT, USA

²Division of Otolaryngology, The University of Vermont Medical Center, Burlington, VT, USA

³Department of Pathology and Laboratory Medicine, The University of Vermont Medical Center, Burlington, VT, USA

Corresponding Author:

Nicole Wershoven, Division of Otolaryngology, The University of Vermont Medical Center, West Pavilion 4, 111 Colchester Avenue, Burlington, VT 05401, USA.

Email: nicole.wershoven@med.uvm.edu



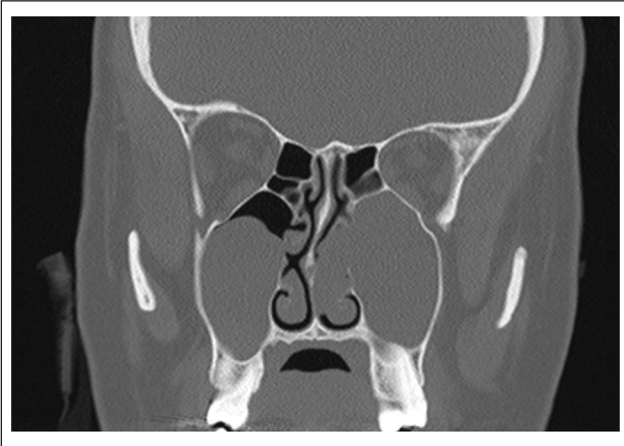


Figure 1. CT scan demonstrating opacification of the maxillary sinuses bilaterally, extending into the nasal cavity on the left.

and mucopurulent nasal drainage. Prior to evaluation, she had been treated with 2–3 rounds of oral antibiotics and steroid nasal spray twice daily with improved symptoms on the right but continued to have persistent symptoms on the left. She denied any previous nasal injuries. Allergy testing was previously negative but she endorsed spring and fall allergy symptoms. Clinical history was additionally negative for asthma.

On nasal endoscopy, she was noted to have a septal deviation to the right. The right middle meatus was narrow but patent with some edema but no frank polyposis or drainage. The left middle meatus was obscured by a polyp. The remainder of her head and neck examination was normal. Computed tomography (CT) of the sinuses demonstrated opacification of the maxillary sinuses bilaterally (Figure 1), extending into the nasal cavity on the left suggestive of antral polyp. She declined treatment with an oral prednisone taper. The patient underwent septoplasty, bilateral maxillary antrostomy with removal of diseased tissue, and inferior turbinioplasty. Intraoperative findings were notable for cysts filled with yellow serous fluid emanating into the middle meati bilaterally from the maxillary sinuses. Final pathology revealed large aggregates of proteinaceous material resembling fibrin with cholesterol clefts, hemosiderin-laden macrophages, and a giant cell reaction consistent with cholesterol granulomas (Figure 2).

Postoperatively, the patient continued nasal saline rinses 2–3 times per day and pain had been managed with ibuprofen and acetaminophen. At her 2 month postoperative visit, nasal endoscopy was concerning for possible recurrence along the anterior margin of the left antrostomy, which was widely patent. Attempted decompression with a curved suction was unsuccessful. She was prescribed a prednisone taper of 40 mg/20 mg/10 mg for 4 days each. She was reevaluated 1 month later and left nasal endoscopy revealed interval growth of the cyst along the anterior/inferior margin of the surgical antrostomy consistent with recurrence of presumed cholesterol granuloma refractory to prednisone. She

underwent revision left maxillary antrostomy with removal of diseased tissue 6 months after her initial surgery. Findings at the time of surgery were consistent with recurrent cholesterol granuloma with the point of attachment identified at the anterior inferior wall. The point of attachment was cauterized intraoperatively. Final pathology was consistent with cholesterol granuloma. Postoperatively she was continued on nasal saline irrigations. She is followed closely but currently has no evidence of recurrent disease 7 months post revision surgery.

Discussion

Cholesterol granulomas can be found in various locations in the body and are a granulomatous entity that forms in response to crystallized cholesterol.⁸ Traditionally, they are found in the middle ear and less frequently so in the paranasal sinuses. The known precipitants of cholesterol granulomas are impaired drainage, poor ventilation, and hemorrhage into the sinus most likely due to nasal trauma or chronic sinusitis.

CT findings usually demonstrate a well-circumscribed, homogeneous mass that may have bony expansion and erosion.¹ They can be isodense or hypodense and mimic several other pathologies.³ For these reasons, microscopic diagnosis is needed and histopathological analysis shows cholesterol clefts admixed with foreign-body giant cells, foam cells, and macrophages that are laden with hemosiderin.⁷ These elements are fixed in fibrous connective tissue and pieces of bone, epithelial residue, and fibrin are other possible elements that could also be present.⁶

A systematic review of the literature from 1970 to December 2010 regarding cholesterol granulomas in the paranasal sinuses was performed by Durgam and Batra.⁹ This review reported findings on a total of 135 patients with cholesterol granulomas of the paranasal sinuses. The mean age was 43.8 years with a male to female ratio of 5.6:1.

In an effort to provide an update to this comprehensive review, we examined the literature regarding cholesterol granulomas of the paranasal sinuses from January 2011 through 2021. Twelve abstracts were identified with a total of the 18 cases. The mean age was 45.4 years with a male to female ratio of 0.7:1.

Combining the results from our review with that of Durgam et al., approximately 59% (91/153) of the reported cases of cholesterol granulomas originated in the frontal sinus, 33% (51/153) in the maxillary sinus, 4% (6/153) in the ethmoid sinus, and 3% (5/153) in the sphenoid sinus (Figure 3).

Given the variability in location and features of cholesterol granulomas in the paranasal sinuses they are amendable to various surgical approaches. Classically, there is the external approach with the Caldwell-Luc operation and an internal approach with endoscopic sinus surgery. Other procedures such as intranasal antrostomy, lateral wall osteotomy, and incisional biopsy of maxillary vestibule were also reported.⁹

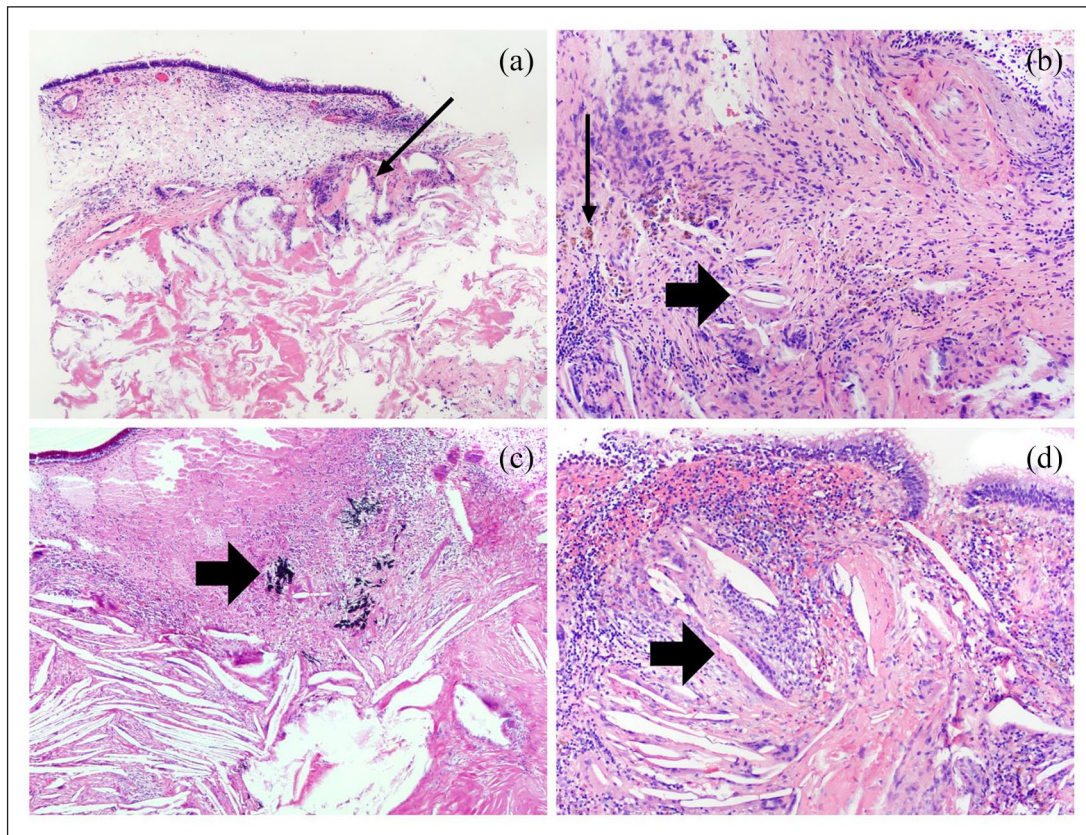


Figure 2. (a) Left maxillary sinus tissue with frequent cholesterol clefts and multinucleated giant cells (long arrow) admixed with amorphous eosinophilic material below the respiratory mucosa and edematous superficial aspect of the submucosa (H&E, 40 \times magnification). (b) Higher power image of the left maxillary sinus cholesterol granuloma demonstrating hemosiderin (long arrow) as well as focal chronic inflammation in addition to the cholesterol clefts and giant cells (short arrow) (H&E, 100 \times magnification). (c) Right maxillary sinus cholesterol granuloma with foci of mineralization (arrow) in addition to the cholesterol clefts surrounded by a rim of chronic inflammation (PAS stain, 40 \times magnification). (d) Higher power image of the right maxillary sinus cholesterol clefts (arrow) with surrounding chronic inflammation below the respiratory epithelium (H&E, 100 \times magnification).

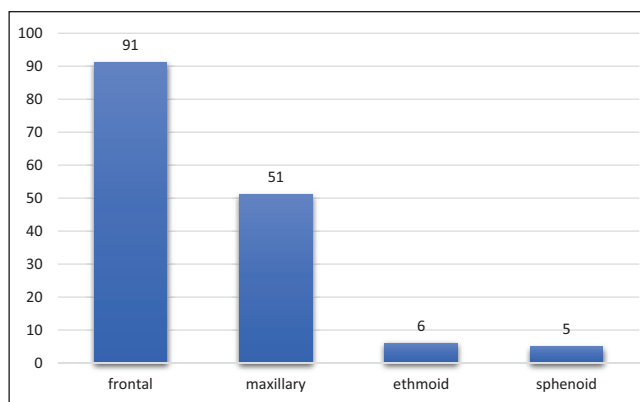


Figure 3. Location of cholesterol granuloma in the paranasal sinuses (n = 153) (1970-April 2021).

The external approach has essentially become obsolete due to the advancements of endoscopic sinus surgery that allow most lesions to be removed endoscopically.¹⁰ The Durgam et al. review reported on definitive management for 65 cases

which consisted of an open procedure in 80% (52/65) of cases and an endoscopic approach in 20% (13/65) of cases. Of the 18 cases reported in the literature since 2011, 61.1% (11/18) were treated with an open approach and 38.9% (7/18) were treated via an endoscopic approach. The Durgam et al. review reported a recurrence rate of 8.2% and in our review of cases since 2011, there were reportedly zero cases of recurrence. Although our patient developed recurrence on the left, she was initially asymptomatic but then developed obstruction and facial/eye pressure several months later requiring revision left maxillary antrostomy.

Interestingly, we describe the second known case of bilateral cholesterol granulomas of the maxillary sinus, with the prior case by Marina and Gendeh.² The patient in the first reported case had 1 year of unremitting nasal blockage and facial pain before undergoing bilateral inframeatal antrostomies followed by sublabial antrostomies.² In contrast to our patient who had no underlying risks for cholesterol granuloma formation, histopathology of the resected mass was consistent with chronic sinusitis and subsequent nasopharyngeal biopsy reported adenoiditis.²

Conclusion

In conclusion, we present the second known case of a bilateral cholesterol granuloma of the maxillary sinus. This case reinforces that upon presentation, cholesterol granulomas can resemble multiple pathologies and tissue for histologic examination is needed for diagnosis. In addition, we illustrate that although rare, cholesterol granulomas should be kept in mind as a possible etiology for bilateral masses of the paranasal sinuses.

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Ethics approval

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Informed consent

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

Presented

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ORCID iD

Nicole Wershoven  <https://orcid.org/0000-0003-0835-2521>

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