

# Sphenoid sinus cholesterol granulomas presenting with abducens nerve palsy

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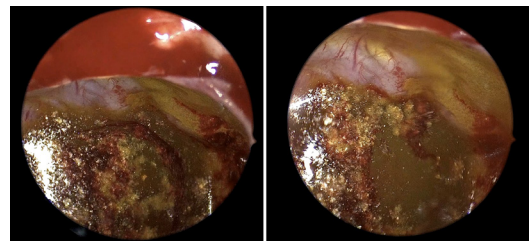
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## SUMMARY

We discuss two patients who presented with sphenoid sinus cholesterol granulomas and associated unilateral abducens nerve palsies. Two case reports from our institution are reported. A literature review of available articles is presented, discussing both cholesterol granulomas of the sphenoid sinus and sphenoid sinus lesions that present with unilateral abducens palsy. A total of nine cases of sphenoid sinus cholesterol granulomas have been previously reported in the literature. A wide variety of sphenoid sinus pathologies can present with unilateral abducens nerve palsies, however no sphenoid sinus cholesterol granulomas with this presentation were found in the literature. In conclusion, cholesterol granulomas should be included in the differential when a patient presents with an expansile mass of the sphenoid sinus and associated unilateral abducens nerve palsy.

## BACKGROUND

Cholesterol granulomas occur most frequently not only in the petrous apex but also in the mastoid bone, middle ear, paranasal sinuses, orbitofrontal bone and petroclival region.<sup>1</sup> Occurrence in the paranasal sinuses is rare,<sup>2</sup> with only nine reported cases to date arising from the sphenoid sinus.<sup>1 3-9</sup> It is also rare for sphenoid sinus lesions to present with isolated unilateral abducens nerve palsy, which occurs due to the immediate proximity of the



**Figure 2** Intraoperative images of cholesterol granuloma within the sphenoid sinus.

cavernous sinus dura to the sphenoid sinus.<sup>10</sup> We report two patients both of whom presented with sphenoid sinus cholesterol granulomas and associated ipsilateral abducens nerve palsy.

## CASE PRESENTATION

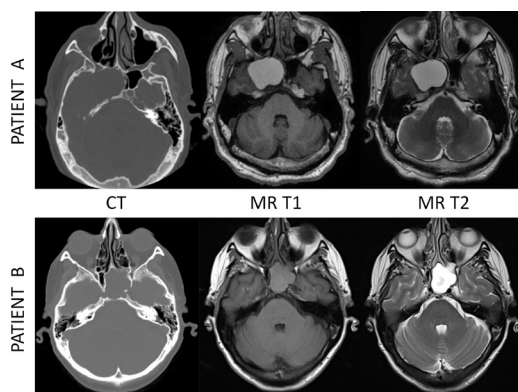
The patients discussed in this case series, although unique, presented similarly. Patient A, a man in his 40s, presented with a 3-month history of right-sided temporal headaches and 10 days of diplopia. Patient B, a woman in her 60s, presented with a 1-month history of headaches and 7 days of diplopia. Neither patient had any other sinonasal complaints. Patient A denied any prior sinonasal trauma or surgery, and patient B only had a remote history of nasal trauma and subsequent septoplasty 30+ years prior to the onset of her current symptoms. Physical examination revealed a lateral gaze palsy towards the right (patient A) and towards the left (patient B), respectively. The rest of their neurological examination was normal.

## INVESTIGATIONS

Nasal endoscopy of the sphenothmoidal recess of both patients was unremarkable. CT scan and MRI were performed for further evaluation. Imaging for patient A revealed a benign-appearing expansile lesion within the right sphenoid sinus, and imaging for patient B showed a benign-appearing expansile lesion located in the left sphenoid sinus. Both lesions showed erosion of the lateral wall, compression of the ipsilateral cavernous sinus and hyperintense signal on both T1 and T2 sequences with a fluid–fluid level (figure 1).

## DIFFERENTIAL DIAGNOSIS

Isolated sphenoid sinus pathology is a relatively uncommon entity. The spectrum of pathology includes inflammatory causes (eg, mucocoeles, fungal ball), neoplasms (eg, inverted papilloma) and other miscellaneous entities (eg, meningoencephalocele).<sup>11</sup>

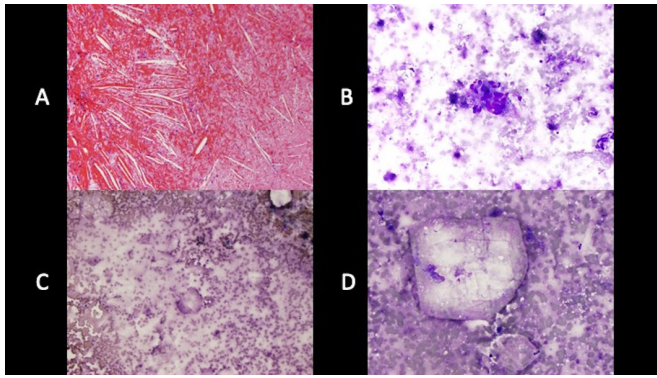


**Figure 1** Top row: patient A. CT scan without contrast demonstrating a mass of the right sphenoid sinus with benign-appearing bony expansion. MRI T1 and T2 demonstrating a sphenoid mass that is bright on both sequences. Bottom row: patient B. CT scan without contrast demonstrating a mass of the left sphenoid sinus with benign-appearing bony expansion. MRI T1 and T2 demonstrating a sphenoid mass that is bright on both sequences.



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**Figure 3** (A) H&E stain at 10× microscopic power of the cystic wall mass. The tissue section demonstrates a fibrous cystic wall with abundant needle-shaped cholesterol clefts and a few irregularly shaped crystals admixed with lipid and haemosiderin-laden macrophages. (B) Diff Quik stain at 20× microscopic power of intraoperative smear preparation of surgically drained cystic content showing multinucleated giant cells in a foamy-lipid background. (C) Diff Quik stain at 10× microscopic power of intraoperative cystic content showing irregularly shaped angulated crystals. (D) Diff Quik stain at 20× microscopic power of intraoperative cystic content showing cholesterol crystals in greater detail.

In order to narrow this differential diagnosis, imaging such as CT scan and MRI are indicated. In our patients, this imaging revealed a benign appearing homogenous expansile mass on CT imaging, with hyperintense signal on both MRI T1 and T2 sequences. Classically, an isolated skull base mass with hyperintense signal on both T1 and T2 sequences is indicative of cholesterol granulomas, however other considerations would include spontaneous organising haematoma or a mucocele with high protein content. The final diagnosis can be supported by intraoperative appearance of the mass and results of pathological examination.

### TREATMENT

Given the new onset cranial neuropathy with worsening symptoms, surgery was performed for both diagnostic and therapeutic purposes. Wide sphenoidotomies were performed (right sided for patient A and left sided for patient B) revealing a cyst contained within the sphenoid and frank cholesterol crystals were visible in yellow-green fluid filling the lesion (figure 2). Pathology obtained intraoperatively confirmed abundant cholesterol clefts, fibrous capsules with embedded inflammatory cells and multinucleated

giant cells in a foamy-lipid background (figure 3). The cysts were widely marsupialised into the nasal cavity.

### OUTCOME AND FOLLOW-UP

Both patients' diplopia and headaches improved significantly in the immediate postoperative period, with complete resolution of diplopia by 48 hours following surgery. Surveillance endoscopy at 6 months postoperatively for both patients showed patent, although partially scarred down, sphenoid ostia.

### DISCUSSION

Cholesterol granulomas are expansile, cystic lesions. Two hypotheses exist for their pathogenesis.<sup>12</sup> Both hypotheses agree that blood enters a mucosalised space, and the cholesterol crystals that result from the anaerobic breakdown of blood create a foreign body giant cell reaction. This results in the formation of a cyst. The first hypothesis proposes that the negative pressure in the mucosalised space is the original cause of the bleeding. The second hypothesis is that exposed bone marrow is the cause of the bleeding, due to an osseous dehiscence between the bone and a pneumatised air cell. Review of both patients presented here did not reveal an obvious origin of the cholesterol granuloma. Both patients' contralateral sphenoid sinuses were unremarkable and their petrous apices were not pneumatised.

While cholesterol granulomas typically occur in the mastoid portion of the temporal bone, the paranasal sinuses are a rare but possible location for these lesions. Durgam and Batra conducted a systematic review in 2012 of cholesterol granulomas occurring in the paranasal sinuses, revealing a total of 135 cases.<sup>2</sup> The most common presenting symptom was related to the orbit, with 89 (66%) reporting retro-orbital pain, visual changes, proptosis or globe deviation. Only three cases (0.2%) were located within the sphenoid sinus. Our updated literature review yielded nine total case reports of sphenoid sinuses cholesterol granulomas, underlining the paucity of this specific location for these cysts.<sup>1 3-9</sup> A summary of the available literature is presented in table 1. Patients presented with a variety of symptoms including compressive optic neuropathy, headaches/migraines, nasal drainage and syncope. Most patients had erosion of the sphenoid sinus due to expansion of the mass. All patients underwent endoscopic resection and had resolution of all or most of their symptoms.

Interestingly, both of our patients presented with headaches and isolated unilateral abducens nerve palsy. Sphenoid sinus masses can present with a variety of cranial neuropathies, including unilateral abducens nerve palsy. This is thought to occur due to the proximity of the cavernous sinus dura to the sphenoid sinus and the expansile features of some masses.<sup>10</sup> There are reports of

**Table 1** Summary of literature discussing sphenoid sinus cholesterol granulomas

Author/s	Age (years)	Sex	Presenting symptoms	Erosion of a sphenoid sinus wall?	Resolution of symptoms after surgery?
Chen and Wang <sup>1</sup>	50	M	Headache, tinnitus, nasal obstruction, nasal discharge	Yes, lateral	Yes
Hwang <i>et al</i> <sup>3</sup>	56	M	Optic neuropathy	Yes, lateral and anterior	Yes
Ahmed <i>et al</i> <sup>4</sup>	48	M	Optic neuropathy	Yes	Not specified
Pehere <i>et al</i> <sup>5</sup>	46	F	Optic neuropathy, headache	No	Yes
Weiland and Aygun <sup>6</sup>	72	M	None	Yes, inferiorly and laterally	Remained asymptomatic
Kang <i>et al</i> <sup>7</sup>	78	M	Facial pain, headache and toothache	Yes, lateral wall	Not specified
Pou <i>et al</i> <sup>8</sup>	30	M	Visual changes, gait instability, headache and nasal congestion	Yes, lateral wall	Yes
Kim <i>et al</i> <sup>9</sup>	67	M	Syncope	Case 1. Yes, only partial	Yes
	49	Not specified	Visual changes, headache	Case 2. Yes, posterior wall and roof	Yes

## Patient's perspective

When asked to give my perspective on this whole sinus thing I thought, 'Do you all really want me to do that? I can be rather chatty without a filter but I will do the best I can'. The best part of the whole thing was the doctor. He did a great job of explaining things to me and calming my nerves from start to the end.

This whole thing started with sinus headaches. Nothing over the counter seemed to help so I just lived with it. (I am the poster child for pain tolerance.) I knew something was wrong when I almost rear ended a vehicle while driving to work one day. I experienced double vision for the first time. It was bad enough that I had to get one of my sons to drive me to work for a week. When it did not clear up in a couple of days it scared me. I immediately thought this cannot be good. There has to be a tumour or something to make me have double vision.

After seeing my general practitioner (GP), he ordered an MRI and it showed there was some sort of mass possibly pressing on my optic nerve in my left sphenoid sinus cavity. He sent me to a neurosurgeon. He told me enough stuff to scare the stew out of me when he explained all the scenarios. He is the one who referred me to the doctor because he thought it was more of an ear, nose, and throat (ENT) problem.

I saw the doctor on a Tuesday and everything was set in motion to get this growth out of my head on Friday (of the same week). Now I was preparing for what I thought was going to be the removal of a tumour that was in close proximity to my pituitary gland and all the things that could possibly go wrong with nicking that during surgery. I was literally prepared to die. Oh yeah, did I mention that we were in the midst of a global pandemic and that the hospital I was having the surgery in was the hospital where they sent people from all over the state with COVID-19 when other hospitals had done all they could for them.

I arrived at the hospital prepared for surgery scheduled for 10:00 trusting that everything was going to be ok. When I woke up in a recovery room, I immediately thanked God that I was alive and then started to see if I was normal (or normal for me) and had control of all my faculties. I was never so happy to have been told that what we thought was a tumour was a cyst, a cholesterol granuloma at that. I was told that it is very rare.

When I sat up in the bed, a bunch of blood and other disgusting stuff came pouring out of my nose. I called the nurse's station for help and they took care of it. I looked surprisingly good for what I was told they did to me. My left nostril felt like a complete football game had been played in it the night before but the pain was not unbearable.

The thing that I was most thankful for was I was not experiencing double vision any more. I was told pre surgery that it may or may not clear up depending on the damage that had been done. I have not experienced any double vision since the surgery either. I still have some drainage, not as much, and not every day. I am ok with that too. If it is draining it does not stop.

Another thing I am thankful for is I did not experience any pain with this surgery. My nose was sore but I think that was to be expected. I did get my prescriptions filled just in case but I threw the pain pills away. I still experience a sinus headache occasionally but this is Louisiana. Everyone has sinus headaches. My smell has also improved. I can smell things now that I had not realised I was not smelling. Do not know if that is psychological or not?

Continued

## Patient's perspective Continued

My life is pretty much back to normal (or as normal as it can be). I am not going to let it slow me down. I have got things to do, places to go and people to see.

## Learning points

- ▶ Sphenoid sinus cholesterol granulomas should be included in the differential when patients present with isolated abducens nerve palsy in the setting of an expansile lesion within the ipsilateral sphenoid sinus.
- ▶ Characteristic imaging findings can include erosion of the lateral wall, compression of the ipsilateral cavernous sinus and hyperintense signal on both T1 and T2 sequences with a fluid–fluid level.
- ▶ Wide surgical drainage is the recommended management of these lesions, and most patients have resolution of their primary presenting symptoms following surgical intervention.

isolated unilateral abducens nerve palsies arising due to sphenoid sinus masses with a variety of pathologies. These include plasma cell leukemias,<sup>13 14</sup> mucocoeles,<sup>15–18</sup> giant cell tumours,<sup>10 19</sup> gastric cancer metastasis,<sup>20</sup> aspergilloma,<sup>21</sup> lymphoma,<sup>22</sup> bronchogenic carcinoma metastasis,<sup>23</sup> primary sphenoid sinus tumours,<sup>23</sup> inverted papilloma,<sup>24</sup> and plasma cell granuloma.<sup>25</sup>

Typical radiographic finding of cholesterol granulomas can be very helpful in narrowing a differential diagnosis. Classically, characteristic MRI findings of cholesterol granulomas are high-intensity signals on both T1-weighted and T2-weighted imaging. This is due to the presence of paramagnetic methaemoglobin accumulation peripherally, from the breakdown of blood products.<sup>2</sup> Characteristic CT findings of cholesterol granulomas include a homogenous, well-circumscribed and opacified paranasal sinus, with accompanying bone erosion.<sup>2</sup>

Classically, the pathology observed in cholesterol granulomas has been described as plasma cells, foamy histiocytes, multinucleated giant cells and cholesterol clefts. Deposited haemosiderin and other blood products are also observed. On gross examination, a cholesterol granuloma consists of a viscous brown or yellow fluid surrounded by a fibrous capsule embedded with inflammatory cells.

Surgical drainage is typically indicated for cholesterol granulomas in order to resolve or improve the symptoms caused by the expansile mass. Careful reviewing of imaging prior to surgery is advised as the expansive properties of the mass often distort anatomy. Considerations during surgery include the proximity of the sinuses to adjacent vital structures such as the optic nerves, the internal carotid arteries and the orbits. Wide openings to the involved sinuses should be achieved as considerable postoperative scarring is expected. Recurrence in the paranasal sinuses is rare and surgery is an effective treatment as most patients have resolution of their primary presenting symptoms.<sup>2</sup>

In summary, we are reporting two unique presentations of isolated lateral rectus weakness and diplopia arising from a sphenoid sinus cholesterol granuloma. While rare, sphenoid sinus cholesterol granulomas should be included in the differential when patients present with isolated abducens nerve palsy in the setting of an expansile lesion within the ipsilateral sphenoid sinus.

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

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