Diagnostic dilemma – sinonasal organizing hematoma

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Abstract

Sinonasal organizing hematomas are benign lesions often mistaken for malignancy due to their aggressive appearance on diagnostic imaging and endoscopic findings that favor advanced disease. The destructive nature of this pathology paired with the rarity of the presentation often results in diagnostic deception that may escalate intervention planning and affect discussion of prognosis with patients. Herein, we present a case of a 56-year-old male with left-sided nasal obstruction and daily epistaxis, where computed tomography imaging revealed heterogeneous opacification of the left maxillary sinus, erosion of the left inferior orbital wall and extension into the nasal cavity. Although clinical and radiographic presentations of sinonasal organizing hematomas can be managed definitively with endoscopic intervention, there is a need to increase awareness of this entity among clinicians to improve our prognostic counseling with patients.

Keywords

Sinonasal organizing hematoma, Sinonasal hematoma, sinonasal malignancy, benign neoplasm, paranasal sinuses, organized hematoma

Introduction

Sinonasal organizing hematomas (SOH) are uncommon, non-neoplastic, hemorrhagic lesions that occur within the paranasal sinuses. SOHs have been reported in the literature as often affecting the maxillary sinus, with few affecting the ethmoid, sphenoid and frontal sinuses. They remain, however, a relatively unknown entity that is often mistaken for malignancy due to their expansive local destruction.1-4 While demographic, clinical, and radiographic findings are useful in the diagnosis of this paranasal sinus mass, immunohistochemical evidence is often necessary to establish a definitive diagnosis.3

Case

A 56-year-old male with history of alcoholic liver cirrhosis presented to a local urgent care for evaluation of 2 months of unilateral nasal obstruction. He was prescribed a course of amoxicillin/clavulanate and instructed to use fluticasone daily. After failing to obtain relief over the next month, his symptoms progressed to include near daily epistaxis, and persistent left-sided periorbital pain and epiphora. After initial consultation with a community provider, where imaging findings on computed tomography (CT) and magnetic resonance imaging (MRI) were noted to be concerning for sinonasal malignancy. He then was referred to our clinic for management of perceived sinonasal carcinoma. During our evaluation, he endorsed unilateral V2 distribution hypoaesthesia, and maxillary pressure, but denied visual changes or dental pain. His left eye demonstrated mild exophthalmos without diplopia. History was notable for prior alcohol abuse resulting in alcoholic liver cirrhosis with clinical thrombocytopenia (platelets 75 x 10³/uL), PT/INR of 15.9/1.3, and hemoglobin level of 33 g/dL. The patient reported isolated epistaxis denying hematemesis or melena other than distant episodes while in acute liver failure.

CT of the sinuses revealed a large sinonasal mass with heterogeneous density in the left maxillary sinus, smooth bony...
destruction of the left medial and lateral maxillary sinus walls, extension into the nasal cavity, and erosion of the left inferior orbital wall (Figure 1(a)). MR imaging with gadolinium revealed a heterogeneously enhancing mass with foci of high T1 and low T2 signals suggesting blood products (Figure 1(b)). Nasal endoscopy demonstrated total obstruction of the left nasal cavity. Biopsy of the encapsulated mass resulted in profound bleeding requiring placement of two Merocels for nasal packing as well as inpatient observation for 24 hours due to persistent bleeding. Initial pathological reports were unable to rule out malignancy, where the specimen was noted to include inflamed respiratory mucosa with organizing thrombus.

Based on these radiologic and clinical findings, we engaged in extensive discussions with the patient and family in preparation for the escalation of care that would be required in the case of malignancy. The patient was consented for resection of the tumor via endoscopic medial maxillectomy, including resection of the inferior turbinate. Early in the resection, a repeat biopsy was sent for frozen pathology, which remained inconclusive in ruling out malignancy. We thus proceeded under the premise of obtaining adequate margins for a presumed malignant sinonasal lesion. The tumor was debulked with mostly compressive findings on adjacent surfaces, other than the anterior inferior maxillary sinus and maxillary roof medial to V2. We dissected up to, but not within the pterygopalatine fossa. We used a 30k drill to polish irregular bony surfaces to arrive at normal appearing bone. The patient was observed overnight for additional bleeding and discharged in the morning.

Final pathology from the lesion was consistent with inflamed respiratory-type mucosa with hemorrhage, fibrin, organizing thrombus, where subsequent staining protocols revealed CD31 marker positivity along small vessels. These findings were consistent and confirmative of a diagnosis of SOH.

Following endoscopic resection, this patient fully recovered with an intact hard palate, other than complete resection of the medial wall. The maxillary cavity appears well mucosalized with no evidence of recurrence at last clinical surveillance 1 year from his date of surgery. The patient denies recurrent epistaxis, no persistent V2 numbness, and no periorbital pressure or pain.

Discussion

SOHs are difficult to diagnose due to their infrequent occurrence and non-specific clinical traits. They are often mistaken for malignant tumors, as in this case, due to their ability to induce profound anatomical changes and local destruction of adjacent tissue planes. Reports of SOHs have steadily increased in the literature since the 2000s, with most affecting the maxillary sinus while demonstrating a predilection for presentation in Asian males. SOHs most commonly present with epistaxis and nasal obstruction, although patients may present with cheek pain and headaches. This is the tenth case of SOH diagnosed in the United States and unique for an individual of Hispanic descent. Whether there exists an ethnic propensity toward those from East Asia or underreporting/misdiagnosis of cases in the literature by clinicians remains unclear.

The etiology of SOHs is currently poorly understood. Accumulation of blood in the maxillary sinus is believed to be the trigger for SOH. Causes of the bleeding into the sinus may include trauma, bleeding diatheses, or a hemorrhagic lesion within the sinus. The negative spiral theory is currently the most accepted theory based on immunohistopathological evidence. Pooling of blood in the paranasal sinuses along with poor sinus ventilation and drainage may lead to hematoma formation. As part of the healing process, necrosis, fibrosis and hyalination occur leading to a capsular
formation, thus preventing reabsorption of the hematoma. Neovascularization develops weak endothelium, leading to epistaxis. The progressive expansion of the hematoma eventually causes demineralization of surrounding structures. SOH has been associated with prior history of head and neck surgery, hypertension, coagulopathies, end stage renal disease and liver cirrhosis.\textsuperscript{1,3} The history of alcoholic liver cirrhosis along with coagulopathies associated with cirrhosis may have contributed to the development of this lesion in our patient. A history of cigarette smoking and atopy have also been described in approximately 11\% and 13\% of those presenting with SOHs respectively.\textsuperscript{2}

CT and MR imaging are critical for diagnosis, although CT imaging may show nonspecific findings.\textsuperscript{7} The aggressive, invasive nature of the orbital and bony walls increase suspicion of a malignant neoplasm. A discerning neuroradiologist, however, will identify imaging findings of an expansive soft-tissue mass, smooth sinonasal wall dehiscence, marked heterogenous signal intensity with a hypointense peripheral rim on T2-weight MR imaging to appropriately diagnoses SOH.\textsuperscript{7,8} Complete endoscopic excision is curative for SOH, although prior to widespread adoption of an endoscopic approach reports in the literature detail the use of Caldwell-Luc, lateral rhinotomy, and Denker’s procedures.\textsuperscript{1}

**Conclusion**

Organized hematomas arising in the sinonasal cavities are an important entity to consider in patients with clinical and radiographic findings that we commonly associate with aggressive neoplasms. The presenting symptoms of unilateral obstruction and epistaxis along with visualization of an expansile mass on endoscopy and imaging immediately raise concern of malignancy. Critical review of the CT and MR imaging, discerning review of the patient’s clinical presentation, and at times tissue biopsy allow for improved pre-operative identification of this benign lesion. Most importantly, only by increasing physician familiarity through case reports and additional research will we be able to improve awareness of SOH as an alternative diagnosis to malignancy. In the above case, had the referring and treating teams been more familiar with SOHs, we may have been able to reduce patient anxiety with appropriate counseling.

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**Ethical approval**

Our institution does not require ethical approval for reporting individual cases or case series.

**Informed consent**

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

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