A Rare Erosive Sphenoid Sinus Organized Hematoma Resulting in Epistaxis and Diplopia: A Case Report and Review of the Literature

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Abstract

Sinonasal organized hematomas (SOH) are rare benign entities that frequently masquerade as malignancies, both clinically and radiographically. They most commonly develop in the maxillary sinus and rarely evolve in the sphenoid sinus. The small capacity of the sphenoid sinus, paired with its anatomic intimacy with the skull base, can portend devastating complications from sphenoid SOH, such as blindness and sentinel bleeding.

We report the case of a 79-year-old male with a remote past medical history of oropharynx carcinoma and renal cell carcinoma, who was actively taking Eliquis for recalcitrant atrial fibrillation. He presented to the Emergency Department with a six-month history of sporadic, yet progressive epistaxis, coupled with 3 months of debilitating fronto-occipital headaches, and one week of worsening diplopia. Urgent MR and CT imaging demonstrated a destructive mass centered in the left sphenoid sinus with erosion of the sella, the clivus, and with early encroachment into the middle cranial fossa.

ENT clinical evaluation with a 30-degree rigid endoscope revealed old blood products and clot emanating from the left sphenoid os. Intraoperatively, a trans-nasal endoscopic wide field sphenoidotomy was performed, revealing extensive congealed blood products with mucosal discoloration and bony erosion of the sphenoid floor. Frozen section pathology demonstrated organized hematoma. No feeding vessel was implicated intraoperatively. Postoperatively the patient had immediate complete resolution of his headaches and diplopia, but persistent, though less severe, epistaxis. A subsequent CTA and cerebral angiogram revealed normal bilateral paraclival and cavernous internal carotid arteries without epistaxis source identification. The patient's dose of Eliquis was reduced and he was started on daily gentamycin nasal irrigations with epistaxis resolution.

With the paucity of reports documenting sphenoid organized hematomas, we review the relevant literature and add our unique case to the skull base annals.

Introduction

Sinonasal organized hematomas (SOH) are unusual, calamitous lesions, that often impersonate malignancies.[1] These neoplasms are thought to evolve from an antecedent hemorrhagic event, followed by content organization, fibrotic change, vascular maturation, with gradual expansion/enlargement over time.[2,3] The evolution of SOH within the sphenoid sinus can be catastrophic due the anatomic intimacy with the dura, cavernous sinus, internal carotid artery, optic nerve, and the pituitary gland.

Six previous cases of sphenoid SOH have been reported in the literature, 4 of which depicted patients presenting with visual sequala.[2-5,10,13] Of these cases, only one subjects vision normalized after surgical intervention.[3] Herein, we report a patient with a history of significant epistaxis, debilitating headaches, and diplopia who was found to have an expansile/destructive mass filling the left sphenoid sinus. Surgical removal revealed organized hematoma and resulted in complete resolution of the patient's cumulative symptomatology.

Discussion

Sinonasal hematomas (SSSH) are rare but potentially significant clinical entities felt to arise from contained intra-sinus bleeding, blood product organization, lack of content resorption, rebleeding, and eventual expansion/destruction.[1,6] These lesions most commonly evolve in the maxillary sinus and rarely are conceived within the sphenoid sinus, with only 6 prior reported cases in the literature.[2-5,13] It has been postulated that these lesions can be caused by direct or indirect craniofacial trauma, surgical or iatrogenic effect, spontaneous bleeding (coagulopathies, vascular malformations, aneurysms), inflammation and infection, and tumor associated hemorrhage.[2,3] Chiang et al. reported a sphenoid SOH in a patient who had undergone previous radiation therapy for nasopharyngeal carcinoma, postulating that RT related tissue effect could also be a culprit in SOH evolution.[8] Recall, our patient had a remote history of adjuvant radiation therapy for oropharynx cancer where the left sphenoid sinus received a cumulative dose of 55 Gy. Anticoagulation has also been invoked as a potential risk factor for SOH formation, a hypothesis potentially supported by our patients' long term Eliquis use.[1,9]

The clinical presentation of sphenoid SOH can be nonspecific, often mimicking other sinonasal pathologies. Common symptoms include headache (retro-orbital or vertex), visual disturbances, epistaxis, and neurologic symptoms due to the anatomic proximity of the sphenoid to the cavernous sinus.[1,5,8] Three prior publications have documented SOH presenting with visual sequelae, 2 with resolution after surgical intervention.[2-4] We add our case to this cohort.

Advanced radiographic imaging is a critical tool in helping establish the diagnosis of sphenoid SOH, distinguishing it from angiomatous polyps, mucoceles, fungal infections, papillomas, and malignancies.[11] Contrast enhanced CT imaging of sphenoid SOH typically demonstrates a heterogeneous, patchy enhancing lesion that can be associated with bony remodeling and or destruction.[11] On MRI, SOH are typically isointense or hyperintense on post contrast T1 weighted images due to hemoglobin breakdown, whereas T2 post contrast weighted images demonstrate distinct heterogeneous signal uptake with a hypointense peripheral shell delineating the fibrous capsule of the hematoma.[11] Angiography can be employed as a component of the work up but is typically reserved for cases where vascular lesions or aneurysms are suspected.[11] Despite distinct radiographic characteristics, it can still be difficult to differentiate between SOH and malignancies, as was the case in our scenario.

Surgical treatment via way of a transnasal endoscopic wide sphenoidotomy is the gold standard therapy for symptomatic sphenoid SOH.[1] Early surgical intervention can result in symptom elimination and the potential reversal of tumor related cranial neuropathies. Anticoagulation reversal, management of contributing vascular anomalies, and rigid control of contributing comorbidities is paramount in preventing recurrence.[9]

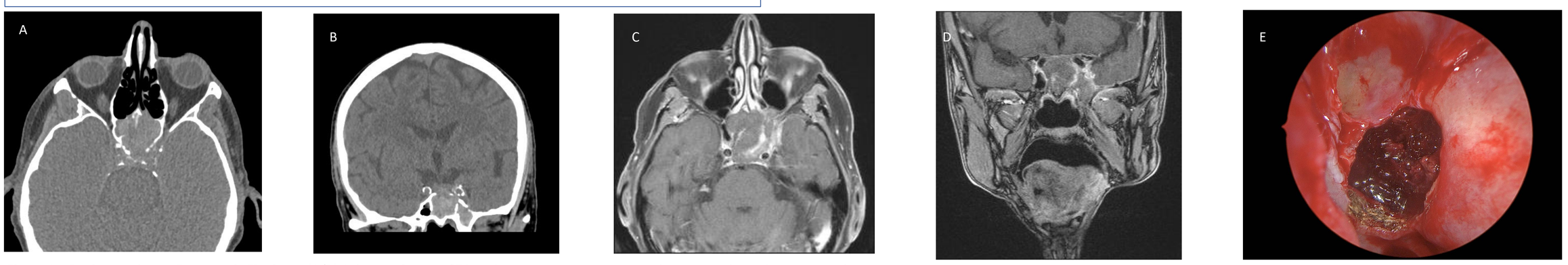

Figure 1 – Radiographic and Endoscopic Findings. (A/B) Axial and Coronal CT imaging demonstrates an expansile, destructive lesion centered in the left sphenoid sinus with erosion/thinning of the walls of the sphenoid sinus, including extension into the adjacent right sphenoid, erosion of the sellar floor, and remodeling of the posterior wall of the upper clivus. (C/D) Contrast enhanced MR imaging illustrates a mild enhancing transpacial sphenoid sinus mass that extends into the middle cranial fossa with focal reactive dural thickening and enhancement. (E) Intraoperatively a wide field sphenoidotomy reveals congealed blood products. (F). A postoperative CT angiogram discloses postsurgical changes consistent with sphenoidotomy, sphenoid opacification resolution, with associated osseous erosion of the left sphenoid wall overlying the left cavernous ICA. (G) A postoperative cerebral angiogram demonstrates normal angioarchitecture of the left ICA and its branches, including the paraclival, paraclinoid, and cavernous ICA.

Case Report

A 79-year-old male presented to the ENT Department for clinical vetting of a newly identified sphenoid sinus mass. The patient, anticoagulated for atrial fibrillation and with a history of oropharynx cancer treated with trimodality therapy, had been suffering from sporadic yet intensifying left sided epistaxis over the previous 6 months. Synchronously he complained of intractable frontotemporal headaches for 3 months which were associated with vomiting and debilitating fatigue. Days prior to his Otolaryngology visit, he was seen in the Emergency room for this constellation of symptoms paired with a one-week history of sudden onset, progressive diplopia. CT imaging demonstrated and expansile, destructive mass/masslike lesion centered in the left sphenoid sinus with erosion of the sella and clivus, and associated anterior, medial, lateral, superior, and inferior sphenoid wall destruction (Fig. 1A,B). A follow up MRI mirrored these findings while also identifying early invasion into the left middle cranial fossa and the left pterygopalatine fossa (Fig. 1C,D).

A 30-degree endoscope was used to exam the bilateral nasal cavities. There was no visually evident mass or lesion, yet blood was noted emanating from the left sphenoid os. An operative transnasal endoscopic biopsy was recommended for diagnostic purposes.

Intraoperatively the patient underwent a posterior ethmoidectomy in conjunction with an extended sphenoidotomy. Coagulated blood products and fibrotic tissue were immediately encountered (Fig. 1E). These contents were completely evacuated, revealing sellar bony erosion with corresponding dural exposure and bony dehiscence over the paraclival carotid. Abnormal appearing mucosa was encountered along the left sphenoid floor, the intra sinus septum, and sphenoid roof. Intrasphenoidal contents and multiple representative soft tissue biopsies were pathologically consistent with organized hematoma and acute on chronic inflammation with abundant necrosis. No dominant feeding vessel was identified as the epistaxis malefactor.

Immediately postoperatively, the patient had complete resolution of his headaches and diplopia, but persistent, though less severe epistaxis over the ensuing 2 weeks. A subsequent CTA and cerebral angiogram revealed osseous erosion of the right sphenoid wall overlying the right cavernous ICA, yet normal bilateral paraclival and cavernous internal carotid artery composition without epistaxis source identification (Fig. 1F,G). The patient's dose of Eliquis was subsequently reduced and he was started on daily gentamycin nasal irrigations with epistaxis resolution. He has had no further bleeds over the ensuing 7 months.

Conclusions

Sphenoid SOH, although rare, should be considered in patients with persistent headache, visual disturbances, or cranial neuropathies, particularly in the context of risk factors such as trauma, surgery, skull base radiotherapy or coagulopathies. Timely radiographic evaluation and surgical management are crucial in minimizing morbidity.

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