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Sphenopalatine Artery Pseudoaneurysm Formation Following Facial Trauma: A case Report and Literature Review

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ABSTRACT

Facial fractures, specifically orbitozygomatic and zygomaticomaxillary complex fractures, are well-documented and common injuries. Pseudoaneurysm formation following cerebrovascular blunt trauma is a rarely experienced complication with an incidence rate of less than 1% with only a few cases reported in the literature. Traumatic pseudoaneurysm formation of the sphenopalatine artery (SPA), the deepest branch of the maxillary artery, is extremely rare due to the deep location of the SPA and its protection from bony landmarks. In craniofacial trauma, pseudoaneurysm formation is not apparent on physical examination due to its deep location and usually presents as persistent nasal bleeding. SPA pseudoaneurysms can present as complications of surgical osteotomies, endoscopic sinus surgeries, facial trauma, or even as a progression of nasopharyngeal cancer. Endovascular embolization provides, safe, quick, and effective treatment while minimizing the morbidity of extensive surgical exposure. In this case report we describe a sphenopalatine artery pseudoaneurysm formation post trauma to provide insight to these rare entities and highlight the importance of early detection and treatment.

1. Introduction

Facial fractures, specifically orbitozygomatic and zygomaticomaxillary complex fractures, are well-documented and common injuries [1,2]. Pseudoaneurysm formation following cerebrovascular blunt trauma is a rarely experienced complication with an incidence rate of less than 1 % with only a few cases reported in the literature [1–6]. Herein, we report our experience with a sphenopalatine artery pseudoaneurysm formation post-traumatic facial injury and perform a literature review to highlight the risk factors, presenting symptoms, and treatment options for such a rare and serious complication.

2. Methods

2.1. Literature search

Using PubMed, Ovid Medline, and Web science, we performed a systemic literature review on April 12, 2022. In all three search engines we started with the search words “sphenopalatine” AND “artery” and “pseudoaneurysm” OR “sphenopalatine” AND “pseudoaneurysm” AND “trauma”. The search became more specific by adding “sphenopalatine” AND “pseudoaneurysm” AND “surgery” OR “endoscopy” AND “complication” AND “facial” AND “trauma”.Duplicates from the different search engines were removed. The inclusion criteria were studies reporting any case of sphenopalatine artery pseudoaneurysm.

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2.2. Literature review and data Extraction:

Articles were screened based on the title and abstract. After that, two authors reviewed the full texts to determine inclusion eligibility and reviewed the references for additional studies that qualified for inclusion. Characteristics collected were year of publication, cause of pseudoaneurysm, presentation, diagnostic modality, and treatment modality.

3. Case report

3.1. Clinical presentation

This is a case of an adult patient who presented to the emergency department in September 2022 after sustaining a right facial fracture. The patient denied any loss of consciousness and physical exam showed a tender lump under the right eye and nasal congestion with epistaxis. On Computed Tomography (CT) scan there was evidence of a right tripod fracture with a fracture of the floor of the right orbit extending through the infraorbital canal and opacification of the right maxillary sinus. (Fig. 1**A**).

During the follow-up period of 2 months, the patient experienced four episodes of epistaxis associated with nasal congestion and pain lasting for 30 min and improving with Afrin-soaked gauze placement. On the follow-up physical exam, the patient had numbness on the cranial nerve V2 distribution on the right side and extreme tenderness on the nasion. The patient does not take any anticoagulation or antiplatelet medication. The patient's brother had Von Willebrand disease however the patient had never been tested for this disease as the patient had never experienced any significant bleeding episodes besides the current epistaxis.

3.2. Diagnosis and imaging

A non-contrast enhanced CT scan of the sinuses was ordered and showed a mass-like obstruction in the right maxillary sinus with obstruction of the osteomeatal unit into the masticator space superior to the right orbit. On further investigation, a computed tomography

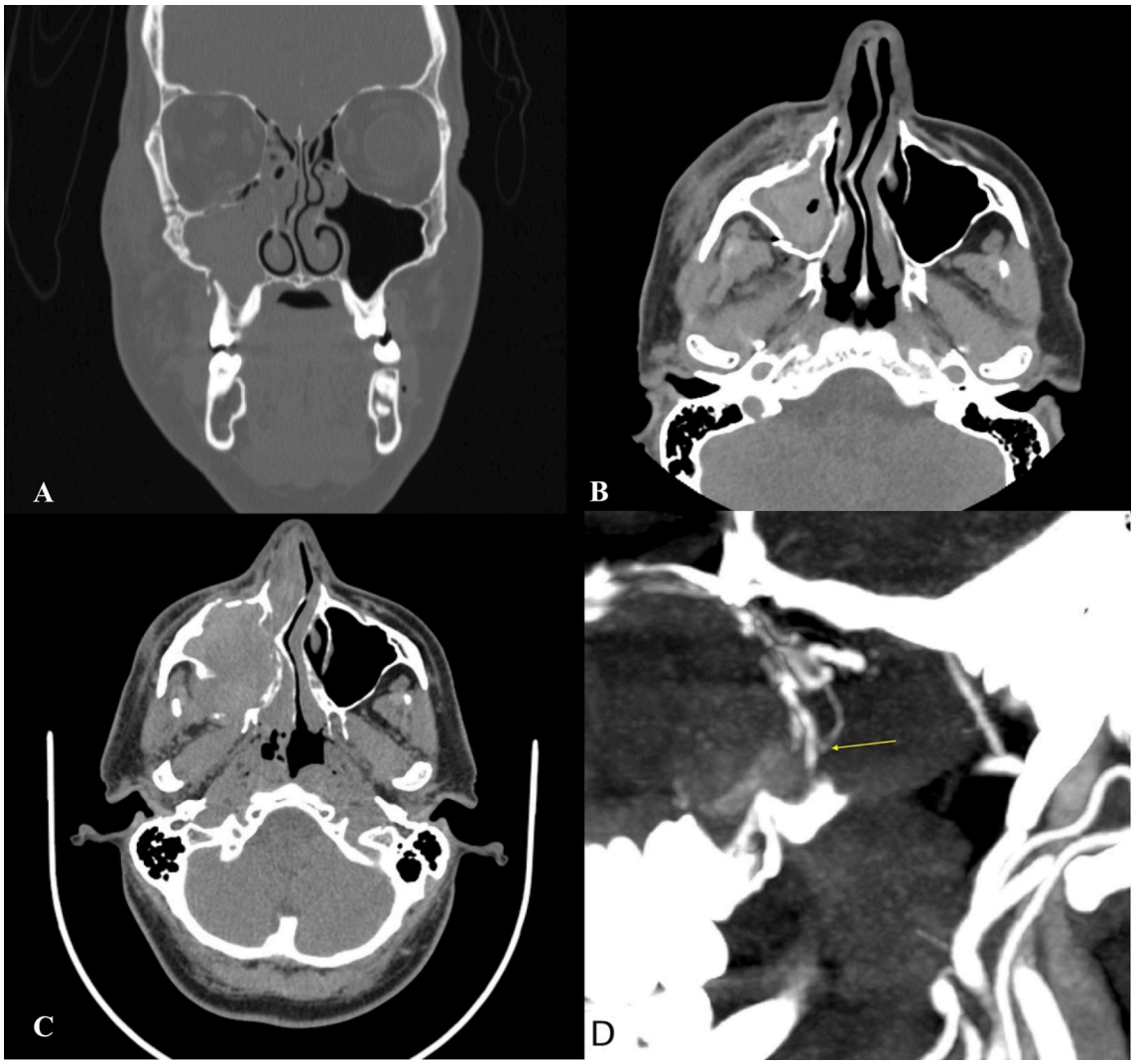

Fig. 1. A. Axial Soft tissue window of initial CT showing the fractured walls of the right maxillary sinus with blood in the maxillary sinus. Note that the retroantral fat pad and region of the pterygopalatine fossa is clear; **B.** Coronal bone window of initial CT showing Zygomaticomaxillary complex fracture on the right with the floor fracture extending through the inferior orbital foramen Axial Soft tissue window of initial CT showing the fractured walls of the right maxillary sinus with blood in the maxillary sinus. Note that the retroantral fat pad and region of the pterygopalatine fossa is clear; **C.** Axial soft tissue window at the same level as 1B of CT 2.5 months later showing progressive expansion of all walls of the right maxillary sinus notably with posterior encroachment on the retroantral fat and on the lateral aspect of the pterygopalatine fossa; **D.** Reconstructed oblique image from a follow-up CTA showing a branch from the Internal maxillary forming a pseudoaneurysm encroaching on posterior maxillary sinus.

angiography (CTA) was obtained and showed an abnormal contrast pooling during the arterial/very early venous phase along the posterolateral margin of the right maxillary sinus just anterolateral to pterygoid. On oblique reconstruction a vessel, presumably from the internal maxillary artery, can be followed directly into the area of abnormal contrast pool highly suggestive of a large pseudoaneurysm. (Fig. 1**B, 1C, 1D**).

3.3. Treatment, hospital course and Follow-up:

The decision to embolize the pseudoaneurysm followed by surgical resection was made based on the otolaryngologist's recommendation seeing that embolization will help in controlling bleeding and decompress the sinus pressure caused by the pseudoaneurysm. The patient underwent a Digital Subtraction Angiography (DSA) which confirmed a right sphenopalatine artery pseudoaneurysm warranting endovascular embolization of the distal internal maxillary artery with 0.6 cc of Onyx-18 (Onyx Liquid Embolic System, Micro Therapeutics, Inc., Irvine, CA) followed by a right -sided antrostomy, sphenopalatine artery cauterization, septoplasty, and clot removal from the right maxillary sinus. (Fig. 2**A, 2B**) Although potential anastomosis with the ophthalmic artery and petro-clival internal carotid artery were not detected with supraselective microinjection, the decision to use liquid embolic instead of coiling was the eliminate the mass effect from the large pseudoaneurysm. The procedure was successful and uneventful. Post-procedure, the patient had normal physical exam and vitals and after an uneventful hospital stay, the patient was discharged home the next day. On the follow-up visit 14 days post-procedure, the patient denied any epistaxis recurrence. The patient underwent post-op Computed Tomography Angiography (CTA), Magnetic Resonance Imaging (MRI), and Magnetic Resonance Angiography (MRA) follow-up angiography which showed normal postsurgical changes. Also, at 2 weeks-post procedure, the patient underwent follow-up MRA, MRI and DSA all showing complete obliteration of the pseudoaneurysm. (Fig. 2**C**).

4. Discussion

Pseudoaneurysm formation is a known consequence of arterial injury, resulting from an incomplete disruption of the arterial wall and forming an expanding lesion with opposing forces from the artery and surrounding tissue [6,7]. Because of this, pseudoaneurysm formation is influenced by arterial blood flow surround tissue elasticity and the extent of the arterial injury [8]. Traumatic pseudoaneurysm formation of the sphenopalatine artery (SPA), the deepest branch of the maxillary artery, is extremely rare due to the deep location of the SPA and its protection from bony landmarks. However, because of its proximity to the pterygoid plates and zygomaticomaxillary complex, pan facial fractures may involve the SPA. Pseudoaneurysm diagnosis in general depends on physical examination which shows a bruit or expanding mass [3,9,10]. However, in craniofacial trauma, pseudoaneurysm formation is not apparent on physical examination due to its deep location and usually presents as persistent nasal bleeding [3–5]. The diagnosis of an SPA pseudoaneurysm is confirmed using a contrast enhanced CT scan or CT angiography (CTA) and the primary treatment of consideration is endovascular embolization [4,5,10,11]. Although surgical ligation is an option, it is not always possible due to the difficulty in reaching deep-lying lesions and the risk of damage to nerves and the internal carotid system [6]. Endovascular embolization provides, safe, quick and effective treatment while minimizing the morbidity of extensive surgical exposure [6,2]. Although in our case the SPA pseudoaneurysm was caused by facial trauma, in other cases SPA pseudoaneurysms can present as complications of surgical osteotomies, endoscopic sinus surgeries, or even as a progression of nasopharyngeal cancer. [12,13].

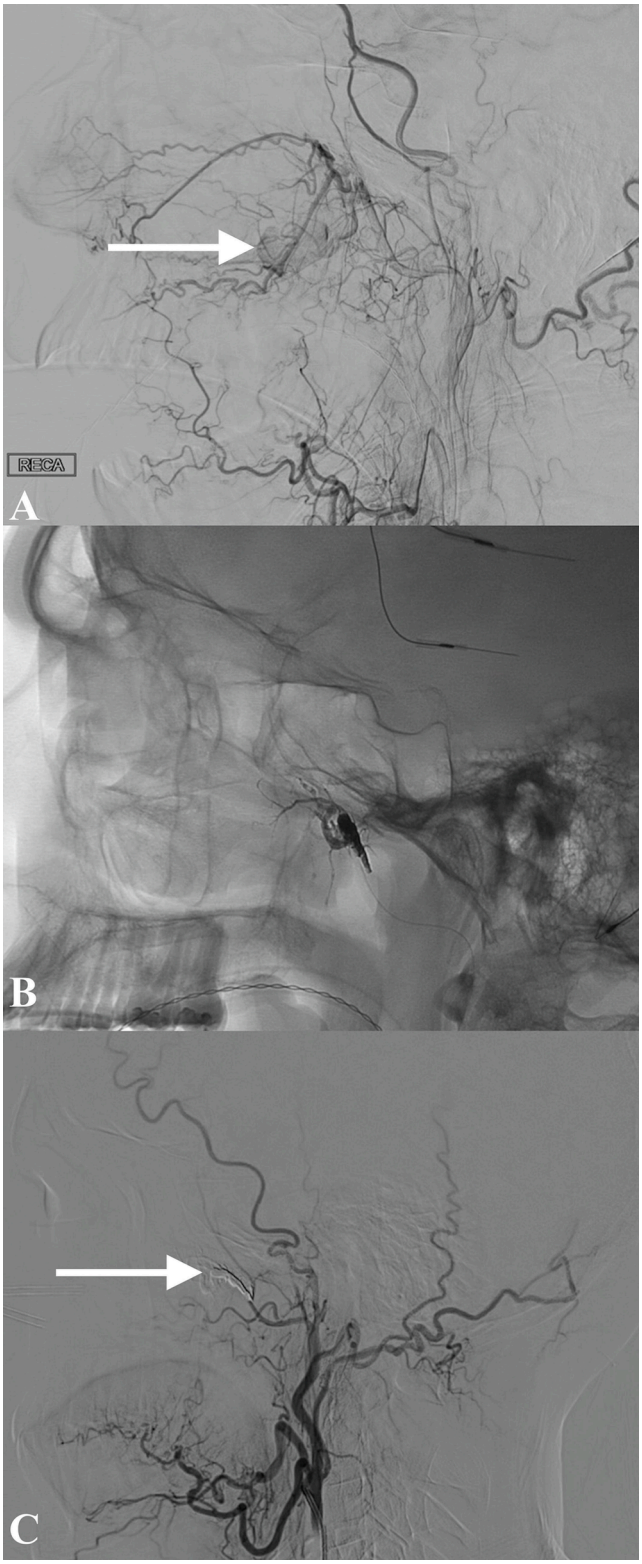

Fig. 2. A. Lateral angiography showing a sphenopalatine pseudoaneurysm. **B.** Fluoroscopy showing onyx embolization of the sphenopalatine artery. **C.** Two-week follow-up angiography showing complete resolution of the pseudoaneurysm.

4.1. SPA pseudoaneurysm following facial trauma

While most of these cases show formation of these pseudoaneurysms due to iatrogenic causes, blunt force trauma and fractures of the

zygomaticomaxillary (ZMC) bone have been demonstrated to cause SPA pseudoaneurysm formation. Early reports of posttraumatic internal carotid artery pseudoaneurysms from Mahmoud et al. described the etiology, prognosis, and management of these rare entities, whereas the understanding of sphenopalatine artery pseudoaneurysms was less understood [14]. Chun et al. presented two cases showing this mechanism of injury as a cause of SPA pseudoaneurysm [2]. Their first case was a patient presenting with a fractured left ZMC following facial trauma. Althrough the bleeding was controlled with cautery and manual compression, recurrent bleeding that traced behind the ZMC and was inaccessible through an open approach and warranted an angiogram which identified an SPA pseudoaneurysm. The pseudoaneurysm was embolized with polyvinyl alcohol particles and coils. Follow-up angiography showed complete obliteration of the pseudoaneurysm. The second case was a patient who presented for a ZMC fracture following blunt trauma to the right malar area. During reduction, there was significant bleeding, so an angiography was performed and demonstrated extravasation through an SPA pseudoaneurysm. Epistaxis was controlled using a gelatin sponge. Follow-up angiography showed complete obliteration of the pseudoaneurysm. Rogers et al. provided a case of a patient who presented following a motor vehicle accident (MVA) with multiple injuries including midfacial soft-tissue and bony injuries [11]. During follow-up, two episodes of recurrent epistaxis that were refractory to nasal packing warranted an angiography which confirmed the diagnosis of a pseudoaneurysm. The pseudoaneurysm was embolized with fiber coils and polyvinyl alcohol particles uneventfully. Moreover, Cohen et al. demonstrated a case of a patient presenting with epistaxis from a pan facial trauma following an MVA [3]. Despite nasal packing, the patient's hemoglobin level continued to decrease which raised suspicion of a distal internal maxillary artery bleed. Angiography was performed and confirmed an SPA pseudoaneurysm which was embolized with detachable platinum coils. The patient's hospital stay was uneventful, and follow-up confirmed unremarkable recovery. Although the mechanism of the SPA pseudoaneurysm formation was similar between these studies and our study, treatment options differed. Because of the large size of the pseudoaneurysm in our study, adjunctive embolization was essential in decompressing the sinus pressure prior to surgical resection.

4.2. SPA pseudoaneurysm following surgical osteotomy

Procopio et al. provided detailed early descriptions of two cases of SPA pseudoaneurysms following orthognathic surgery [15]. The first patient underwent a successful and uneventful surgery, but during follow-up, the patient experienced 6 episodes of recurrent epistaxis resistant to nasal packing and embolization. Surgical exploration was performed and the pseudoaneurysm was clipped definitively. In the second case, the patient had two post-operative episodes of severe epistaxis controlled with anterior nasal packing. CT scan revealed a right SPA pseudoaneurysm which was embolized with complex helical fibered platinum coils uneventfully. Later reports by Kim et al., Maleux et al. and Kumar et al. all provide additional cases showing similar mechanisms [16–18].

4.3. SPA pseudoaneurysm following endoscopic sinus surgery

Campell et al. reported a case of a patient who underwent endoscopic sinus surgery after a CT scan revealed opacification of the left sinuses [19]. Intraoperative bleeding persisted despite monopolar cautery, Surgicel, and rope packing. Subsequent endoscopic examination revealed a pulsatile mass at the left SPA foramen which was then visualized by CTA. Angiography confirmed the diagnosis of an SPA pseudoaneurysm which was embolized with polyvinyl alcohol particles and platinum coils. Gokdogan et al. presented a similar case of a patient with intractable epistaxis resistant to nasal packing following endoscopic sinus surgery for nasal polyposis [12]. Posterior bleeding was observed despite nasal packing. Emergent angiography was performed and identified a right SPA pseudoaneurysm. Embolization was performed with polyvinyl alcohol particles, and the complete obliteration was observed on follow-up angiography.

4.4. SPA pseudoaneurysm as a progression of nasopharyngeal cancer

Additional rarer presentations should also be noted. Gordhan et al presented a unique case of a patient presenting with severe oronasal bleeding with previously diagnosed invasive squamous cell carcinoma of the left alveolar ridge, left nasal cavity, and maxillary sinus treated with]gross total resection, radiation and chemotherapy [13]. Control of the bleeding was attempted using epinephrine and direct pressure but failed. Angiography of the left external carotid artery identified a fistulous pseudoaneurysm connecting the left sphenopalatine artery and the pterygoid plexus, which was embolized using *n*-BCA in ethmoidal and tantalum powder. Complete obliteration was observed with immediate cessation of bleeding and no complications. Table 1 provides a summary of the literature.

Based on our systemic review, we suggest a few key findings that should be taken into consideration. First, the main presenting symptoms of SPA pseudoaneurysms is epistaxis which may be overlooked as nasal bleeding is common after facial injury or surgery. Second, bleeding can occur in distal branches of the maxillary artery that enter the pterygoid plate. Therefore, any bleeding that persists after facial trauma or surgery and is not controlled by nasal packing or other forms related to the protocol for controlling hemorrhage after facial trauma should raise suspicion of an SPA pseudoaneurysm and warrant further investigation with imaging modalities such as CT scans CTA's and DSA's [20]. DSA is the most diagnostic modality since it provides further detail about intraaneurysmal thrombosis or any ongoing hemorrhage. Treatment options should include the endovascular approach either as the primary treatment option or as an adjunct to surgery as it provides immediate control of bleeding and minimal recurrence risk. The decision to embolize the pseudoaneurysm followed by surgical resection in our case report was made based on the otolaryngologist's recommendation seeing that embolization will help in controlling bleeding and decompress the sinus pressure caused by the pseudoaneurysm before surgery. Lastly, close radiological follow-up is recommended as pseudoaneurysms have a high tendency of recurrence [21,22].

5. Conclusion

SPA pseudoaneurysms are rare entities and may develop because of facial trauma, surgical and endoscopic interventions, or as a progression of nasopharyngeal tumors. A low threshold for diagnosing and treating such entities is essential for controlling the hemorrhage and preventing morbidity and mortality. Endovascular embolization provides immediate blood control should be included as a primary treatment option or as an adjunct to the surgical approach.

Disclosures

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Data sharing statement: The relevant anonymized patient-level data are available on reasonable request from the authors.

Declaration of Competing Interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: Pascal Jabbour reports a relationship with Medtronic that includes: consulting or advisory. Stavropoula Tjoumakaris reports a relationship with Medtronic that includes: consulting or advisory. Stavropoula Tjoumakaris reports a relationship with MicroVention Inc that includes: consulting or advisory. Pascal Jabbour reports a relationship with MicroVention Inc that includes: consulting or advisory. Michael Gooch

Table 1
Summary of literature pertaining to sphenopalatine artery pseudoaneurysm presentation, diagnosis, and treatment.

Author	Publication Year	Cause of Pseudoaneurysm	Presentation	Diagnostic Modality	Treatment Modality
Chun et al.	2019	Facial Trauma	Epistaxis	Surgical Exploration and DSA	Manual compression, embolization with polyvinyl alcohol particles, gelatin sponge, and coils
		Facial Trauma	Epistaxis	DSA	Embolization with gelatin sponge
Rogers et al.	1995	Facial Trauma	Epistaxis	DSA	Embolization with coils and polyvinyl alcohol particles
Cohen et al.	1999	Facial Trauma	Epistaxis	DSA	Coil Embolization
Procopio et al.	2003	Surgical Osteotomy	Epistaxis	CT Scan followed by DSA	Coil embolization x2, clipping
		Surgical Osteotomy	Epistaxis	CT Scan followed by DSA	Coil embolization
Kim et al.	2013	Surgical Osteotomy	Epistaxis	CT Scan followed by DSA	Embolization with lipiodol and *N*-butyl-2-cyanoacrylate
Maleux et al.	2019	Surgical Osteotomy	Epistaxis	CT Scan followed by DSA	Embolization with ethiodised oil and *n*-butyl-cyanoacrylate
Kumar et al.	2021	Surgical Osteotomy	Epistaxis	CTA	Embolization with gel foam x2
Campbell et al.	2012	Endoscopic Sinus Surgery	Epistaxis	CT Scan followed by DSA	Embolization with polyvinyl alcohol particles and platinum coils
Gokdogan et al.	2014	Endoscopic Sinus Surgery	Epistaxis	DSA	Embolization with polyvinyl alcohol particles
Gordhan et al.	2013	Progression of Cancer	Epistaxis	DSA	Embolization with *n*-BCA in ethmoidal and tantalum powder
Current Case	2022	Endoscopic Sinus Surgery	Epistaxis	CT Scan followed by CTA, followed by DSA	Endovascular Onyx embolization followed by antrostomy, septoplasty, and clot removal

reports a relationship with Stryker that includes: consulting or advisory. NA.

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