

CASE REPORT

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Delayed post-traumatic intractable epistaxis due to pseudoaneurysms in concha bullosa: a case report

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Abstract

Background Delayed post-traumatic bleeding at various sites is known due to vascular abnormalities related to structural changes in the vessel walls, in the form of pseudo aneurysms and arteriovenous malformations. Delayed epistaxis following facial trauma is a distinct entity that presents as recurrent episodes of bleeding from the nose. History of trauma is important to differentiate it from idiopathic intractable epistaxis, the management of which is less dramatic in the beginning in the form of endoscopy and electrocauterization. However, uncontrolled post-traumatic delayed epistaxis usually requires embolization. We report a rare case of intractable epistaxis associated with multiple pseudoaneurysms (PA), due to bleeding inside concha bullosa, where even embolization was unsuccessful.

Case presentation A 25-year-old male patient presented to casualty and was referred to the Otorhinolaryngology department for recurrent profuse bleeding from left nasal cavity, where conservative measures like nasal packing, and electrocauterization did not help. Patient had a history of fall from height with facial bone fractures 1 month back. Angiography revealed blush with dilatation (pseudoaneurysms) in the nasal branches of internal maxillary arteries, ascending pharyngeal and labial arteries and embolization was done in the Interventional Radiology Department. Post-embolization, the patient again developed massive epistaxis. The patient was explored under general anaesthesia, to find out the site of bleed by nasal endoscopy. Collected blood was found in the left concha bullosa. Conchoplasty of the left middle turbinate was done with cauterization of bleeding vessels. The patient had no further epistaxis and was discharged in stable condition. He is asymptomatic for the last 6 months.

Conclusion Post-traumatic pseudoaneurysms is a rare cause of delayed epistaxis following nasal trauma. Angiography helps in the localization of the source of bleeding inside nasal cavity and can identify pseudoaneurysmal dilations of vessels. Our case highlights the importance of hidden bleeding points inside concha bullosa as a source of intractable epistaxis unresponsive to embolization.

Keywords Epistaxis, Pseudoaneurysm, Embolization, Nose

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Background

Delayed post-traumatic bleedings at various sites are known due to acquired vascular abnormalities related to structural changes in the vessel walls in the form of pseudoaneurysms (PA) and other rare entity arteriovenous (AV) malformations [1, 2]. Delayed epistaxis following facial trauma is a distinct entity that presents as recurrent episodes of bleeding from the nose. History of trauma is important, otherwise, it may be considered as idiopathic intractable epistaxis, the management of which is less dramatic in the beginning in the form of endoscopy and electrocauterization [3]. It is necessary to differentiate post-traumatic intractable epistaxis from idiopathic intractable epistaxis. Pseudoaneurysms, which occur as a result of trauma, as complications of vascular access surgeries, prolonged vascular intervention, and use of antiplatelets or anticoagulants all these may cause the full thickness of the arterial wall to rupture, allowing blood to spill into the surrounding tissue and causing a pulsating hematoma with the perivascular connective tissue compressing and forming a sac. Post-traumatic pseudoaneurysms can arise from both branches of internal and external carotid artery, especially the internal maxillary artery, cavernous internal carotid artery (ICA), and have been reported following maxillofacial trauma [4]. Intractable epistaxis patients frequently seek treatment in the emergency room. Conservative treatments including pressure, nasal packing, and medicines like oxymetazoline help stop bleeding. Surgical intervention may be necessary for intractable epistaxis. In difficult-to-reach surgical areas, embolization can be used to control and reduce the risk of bleeding and revascularization of the pseudoaneurysm by collateral circulation [5].

We present such an interesting case that required both endovascular management and surgical management in the form of conchoplasty for intractable bleeding. Most probably the first to encounter multiple pseudoaneurysms in small vessels of the nasal cavity and inside the concha bullosa. Such a cause of intractable epistaxis has not been previously reported.

Case presentation

A 25-year-old male patient presented to casualty and was referred to the Otorhinolaryngology department for a profuse left nasal cavity bleed for which he had received gel foam nasal packing at a nearby hospital. When he presented in casualty at our hospital, there was no active nasal or oral bleeding. The patient had experienced 2–3 similar epistaxis episodes that stopped spontaneously in the last 1 month. Before 1 month, the patient had a history of fall from height (2nd floor of a building) due

to which he sustained multiple injuries in the form of Fracture of the bilateral anterior wall of maxillary sinus and fracture of lesser wing of right sphenoid, which was managed conservatively and fracture of right femur. Immediately after the fall, the patient had a history of nasal bleeding completely soaking two small face towels, and stopped spontaneously. The patient was managed conservatively for nasal bleeding at that time. For the fracture of the femur, he received open reduction and internal fixation. The patient did not give any history of diabetes, hypertension, tuberculosis, bleeding diathesis, or any other systemic illness. There was no family members any genetic disorders.

At the time of admission in our hospital, on general examination, pulse rate and blood pressure were within normal range, and systemic examination did not reveal any abnormality. Haemoglobin at the time of admission was 10.8 gm%, and complete blood count, urine examination, liver function tests, kidney function tests, coagulation profile, fasting, and postprandial blood sugar, were within normal limits; and ELISA for HIV, HCV, and HBsAg was negative. After pack removal, nasal endoscopy was done and trichloroacetic acid (TCA) application was done over the small abrasion present on the anterior part of the septum which was suspected initially to be source of epistaxis. Also, there was a crust over the left middle turbinate, no other abnormalities were present in the nose and oral cavity. Betadine-soaked gel foam was placed in left nostril at the level of the middle turbinate. However, the patient experienced sudden onset of nasal bleeding on next day and again nasal packing was done. The bleeding stopped on anterior nasal packing. After 2 days the patient was posted for pack removal in Operation Theatre. The bleeding point was identified over the left concha bullosa (pneumatized middle turbinate) which was electrocauterized under local anesthesia. There was no active bleeding after pack removal. Medicated Gel foam was placed in nasal cavity. The patient had an episode of sneezing and again experienced profuse nasal bleeding, which was approximately 500 ml in amount this time. Bleeders could not be identified because of the severity of the bleed and nasal packing was done again which was of temporary help and the profuse nasal bleed started again. This time, the Patient was taken for emergency angiography and embolization was done in the Interventional Radiology department. Angiogram was done and revealed blush with dilatation (pseudoaneurysm) in the nasal branches of internal maxillary arteries, ascending pharyngeal and labial arteries (Fig. 1).

Selective cannulation of the arteries was done. Poly Vinyl Alcohol 300 particles were infused until reduction in the blush and stasis was received. Post-embolization



Fig. 1 Angiogram showing blush with dilatation (pseudoaneurysms) in the nasal branches of maxillary arteries, ascending pharyngeal, and labial artery

angiography revealed a significant reduction in blush in the nasal branches of labial, internal maxillary, and ascending pharyngeal arteries (Fig. 2).

Post-embolization, the patient was given blood transfusion following which he developed massive epistaxis with approximately 500 ml blood loss. Again re-packing was done, and with each episode of bleeding, the hemoglobin fell from 10.8 gm% to 6.9 gm% to 5.3 gm% to 3.4 gm% over a period of 12 days. The patient was then shifted to the medical ICU and investigated for “transfusion reaction”. Other hematological investigations, Peripheral smear, Coagulation profile, and Coombs and Sickling test was performed which came

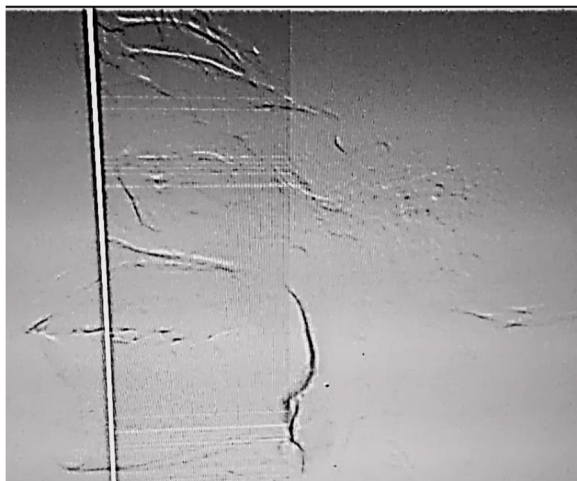


Fig. 2 Post-embolization angiography revealed a significant reduction in blush in the nasal branches of labial, internal maxillary, and ascending pharyngeal arteries

out to be negative. The patient was transfused 1 unit of whole blood, and 4 units of packed red blood cells. This time pack was removed gradually over a few days. Haemoglobin reached a level of 8.4 gm%. The patient was asymptomatic for the next 7 days. He had another massive epistaxis when discharge was being planned.

The patient was taken to Operation Theatre and explored under General Anaesthesia, to find out the site of bleed by nasal endoscopy. Bleeding point was found over the anterior aspect of middle turbinate and was not controlled even after electrocauterization. Collected blood was found in the left concha bullosa (Fig. 3). Conchoplasty of the left middle turbinate was done with cauterization of bleeding vessels inside concha bullosa.

The patient had no further epistaxis after this and was discharged in stable condition. He is asymptomatic for the last 6 months.

Discussion

Post-traumatic pseudoaneurysm is characterized by delayed, increasingly severe, and sometimes fatal epistaxis. The average time between trauma and pseudoaneurysm rupture is approximately 3 weeks [6]. An aneurysm is an abnormal widening or ballooning of a portion of a blood vessel caused by a ruptured blood vessel. A true aneurysm involves all three vessel layers. A false or pseudo aneurysm caused by trauma is characterized by partial vessel transection and hematoma formation, manifesting as an outpouching [7]. Another entity is post-traumatic AV malformation, which is very rare but does not cause spontaneous bleeding. Traumatic arteriovenous malformations (AVM) are typically complicated abnormal communications between arteries and veins. They are most prevalent in the extremities. The trauma that results in an AVM may be penetrating, blunt,



Fig. 3 Collected blood was found in concha bullosa on left side

postoperative, or inflammatory. The main presenting symptoms are facial edema, pulsatile tinnitus, throbbing headache, and bleeding. Doppler ultrasonography and selective arteriography will provide the necessary diagnostic information. Embolization therapy may be a useful adjunct to surgery or the sole treatment if surgery cannot be performed. Proper identification of the feeder's vessels, surgical control of these vessels, complete excision of the AVM, and functional and cosmetic reconstruction of the tissue defect continue to be the basis of treatment [2, 8]. Only a single case of nasoseptal post-traumatic AV malformation was reported following fall which was managed conservatively [9]. Pseudoaneurysm (PA) is a rare life-threatening complication that consists of an incomplete tear of a large blood vessel, causing extravasation of blood into the surrounding tissues. They can result in hematoma formation, in which surrounding tissues are inelastic [1, 4, 5, 10]. Post-traumatic pseudoaneurysm following maxillofacial trauma can occur in the internal and external carotid artery and its branches [11]. Although small ICA pseudoaneurysms can be asymptomatic, they can occasionally manifest as massive epistaxis. Due to the fact that the cavernous portion of the ICA indents the lateral wall of the sphenoid sinus, nasal bleeding can be a symptom of a cavernous ICA aneurysm. Even though ICA pseudoaneurysms are uncommon, they must be considered in the differential diagnosis of intractable epistaxis, especially if a history of maxillofacial trauma is present. A ruptured ICA pseudoaneurysm has a high mortality rate; early diagnosis and treatment can save lives [12]. Pseudoaneurysm of the internal maxillary artery due to a traumatic event has been reported. An abrupt movement of the artery in a "concussion fashion" might cause the early artery lesion to occur. This technique can account for instances where the entity of fractures does not appear to be sufficient to directly result in an arterial lesion. In particular, in those tracts that are more constricted by dense connective tissues, the hematoma might form a PA due to the inelasticity of the MA's surrounding tissue [10]. Intractable epistaxis due to pseudoaneurysm in nasal arteries in concha bullosa, with delayed onset after trauma, has not been reported previously. The likely mechanism for this appears to be that sudden concussion due to trauma may cause abrupt movement of the small branches of vessels in the nasal cavity, with the partial shearing of the vessel wall leading to its weakening and dilatation, and formation of pseudoaneurysm. The presence of concha bullosa and lack of external compressing effect on the vessels may be a cause of delayed bleeding, as vessels start rupturing due to progressive thinning of vessel wall, and lead to profuse intractable and recurrent epistaxis.

Various protocols for the management of Idiopathic Intractable epistaxis have been evolved which comprise of transnasal endoscopic sphenopalatine ligation, transantral ligation of internal maxillary artery, and some favor embolization as primary treatment modality. However, since epistaxis following trauma can be due to multiple pseudoaneurysms, as in our case, and arise from branches of both Internal and external carotid arteries, other than internal maxillary artery, endovascular procedures may fail and more dramatic measures may be needed like internal or external carotid artery ligation or clamping as last resort. Pseudoaneurysms are treated by various surgical and endovascular options [13]. Site of bleeding identified by angiography like in internal carotid artery, internal maxillary artery, and cavernous ICA. Metallic coils, absorbable sponge gel, polyvinyl alcohol particles, n-butyl cyanoacrylate, and polymers are various agents that are used for embolization [14]. After nasal packing and/or cauterization as first-line treatment, embolization may be considered; however, nasal endoscopy should be attempted whenever possible to identify the epistaxis site. Nasal endoscopy can be performed while the patient is under local or general anesthesia; it identifies the potentially offensive region and should be performed routinely prior to embolization and/or vessel ligation [15]. This is highlighted by our case where bleeding vessels were present inside Concha Bullosa.

In the presence of multiple pseudo aneurysms at the end arteries, they are difficult to embolize and if the main trunk is embolized then the chance of recurrent bleeding is high. Therefore, exploration under general anesthesia, to find out the site of the bleed by nasal endoscopy is necessary. Electrocauterization of the bleeding vessels with pseudoaneurysmal dilatations, was performed after opening the concha bullosa. Conchoplasty in our case not only facilitated the visualization of the bleeding vessels, but also in removing the air around the pseudoaneurysms, which could be aggravating or perpetuating the pseudoaneurysms.

Conclusion

This is a case of post-traumatic intractable epistaxis due to hidden bleeding points inside the concha bullosa as a source of bleeding. The angiogram revealed pseudoaneurysm of the nasal branches of internal maxillary arteries, ascending pharyngeal and labial arteries and with initial failed endovascular management. Lack of compression due to air in a pneumatized middle turbinate was the cause of recurrent bleeding in our case. Conchoplasty with electro cauterization of bleeding vessels resulted in remission of condition. Our case

highlights the importance of hidden bleeding points inside concha bullosa as a source of intractable epistaxis unresponsive to embolization.

Any patient with post-traumatic intractable epistaxis should preferably undergo early angiography in each case to identify the bleeding source in form of pseudoaneurysms or AV malformations. This should be followed by surgical exploration under general anaesthesia, according to the likely site of bleeding and cauterization, ligation, or clipping of the offending vessels. Embolization may not be effective in such cases.

Abbreviations

PA	Pseudoaneurysm
TCA	Trichloroacetic acid
AV	Arteriovenous
AVM	Arteriovenous malformations
ICA	Internal carotid artery

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Authors' contributions

SJ and MM prepared the manuscript. SJ revised it critically for important intellectual content. VH contributed towards data acquisition and approved the final draft of the manuscript. AR contributed towards data acquisition and approved the final draft of the manuscript. RS contributed towards data acquisition and approved the final draft of the manuscript. PB contributed towards data acquisition and approved the final draft of the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials

All data sets collected during this study are available upon reasonable request from the corresponding author.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent was taken from the patient.

Competing interests

The authors declare that they have no competing interests.

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