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Case Report

Massive recurrent epistaxis in traumatic pseudoaneurysm of sphenopalatine artery: Report of 2 cases a,aa

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ABSTRACT

Massive recurrent epistaxis because of traumatic pseudoaneurysm of the sphenopalatine artery is rarely diagnosed. It is formed by partial rupture of the endothelium by facial trauma and managed with embolization and maxillofacial reconstruction. Here, we report a case of 2 massive recurrent epistaxes and 1 case of carotid cavernous fistula, which needed embolization to control the recurrent bleeding following trauma. Epistaxis is first managed by tampon, then referred as needed for endovascular intervention by sphenopalatine artery embolization and followed by maxillofacial reconstruction. Recurrent epistaxis was successfully treated with endovascular embolization.

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Introduction

Epistaxis is defined as a flow of blood from the nasal fossae. It is common and usually benign; the great majority of cases do not require medical care [1]. However, some cases can be a surgical emergency because of the amount of blood, repeated episodes, other accompanying symptoms, or the cause [2]. Epistaxis is thought to be idiopathic or essential in 70% of cases; other causes include trauma, surgery, tumors, and malformations [3,4].

Fracture of the zygomaticomaxillary complex is common in facial fractures, but few reports exist of pseudoaneurysm in facial fractures. Pseudoaneurysm, which can occur after partial rupture of the blood vessel wall, has a much higher risk of rupture than true aneurysm malformations [5,6].

Although rare, endoscopic arterial embolization is the treatment of choice for severe, recurrent epistaxis or other forms of hemorrhage. It is effective and increasingly popular as the main treatment [3,7,8]. In this case series, we report 2 cases of massive recurrent epistaxis after facial fractures. The mainstay treatment was embolization and maxillofacial reconstruction after angiogram because of the recurring epis-

taxis that was overlooked by surgeons. This case has been reported in line with the Updating Consensus Surgical CAse REport (SCARE) Guidelines [9,10].

Case presentations

Case 1

A 24-year-old male patient was transferred to our general hospital for loss of consciousness because of a traffic accident 5 hours prior. The patient presented with an anterior skull base fracture, dentoalveolar fracture, and Le Fort type 2 fracture (Fig. 1). He was conservatively treated and discharged uneventfully. The following week, the patient presented with massive recurring epistaxis that was stopped with tampon insertion. He was hospitalized twice with the same problem. Thus, the patient was scheduled for further examination in which angiography and maxillofacial reconstruction were performed. The angiography showed a pseudoaneurysm of the right sphenopalatine artery, on which embolization was performed (Fig. 2). After the embolization and maxillofacial



Fig. 1 – A preoperative computed tomography scan of the facial bones in a 24-year-old male patient presenting with dentoalveolar fracture, Le Fort type 2 fracture, and anterior skull base fracture following blunt trauma.



Fig. 2 - Angiography of the patient revealing a pseudoaneurysm of the right sphenopalatine artery (arrow).

reconstruction, no additional bleeding was found, and the patient was discharged with postoperative follow-up.

Case 2

A 24-year-old male patient presented to our emergency room with complaints of headache and massive recurrent nasal bleeding from the left nose since 6 hours prior. The patient had a history of nasal fracture, bilateral zygomatic fracture, mandibular parasymphisis fracture, and conservative epidural hematoma treatment 2 weeks prior because of a traffic accident (Fig. 3). He had been scheduled for maxillofacial reconstruction, then presented with the emergent complaint; thus, angiography was performed. The angiography demonstrated another extravasation from a pseudoaneurysm of the left sphenopalatine artery (Fig. 4). The patient was treated with embolization and maxillofacial reconstruction. No complication or recurrence of the epistaxis was detected even after the postoperative follow-up.

Discussion

Epistaxis, a flow of blood from the nasal fossae, is a common manifestation after a head injury. In most cases, it is benign and occurs because of a laceration of the nasal mucosa or tear of the anterior ethmoid or sphenopalatine artery after the head injury [1,11,12]. A facial fracture that contributes to trauma of the sphenopalatine artery may also lead to the formation of a pseudoaneurysm. A pseudoaneurysm is an abnormal dilatation of a vessel that leads to an expanding lesion between the vessel and the surrounding tissues. After the disruption of the vessel wall, hemorrhage occurs into adjacent tissues until the pressure of the formed hematoma counterbalances the pressure flow, which compresses the defect and stabilizes the bleeding. However, pseudoaneurysm formation from the sphenopalatine artery is extremely rare because its small caliber and deep location in the mid-face have proven to be protective from blunt and superficial penetrating injuries. The rarity of sphenopalatine artery pseudoaneurysm is due to the small size of the branches of the external carotid artery, which makes a complete cut more likely than a partial laceration [11,13].

The diagnosis of a pseudoaneurysm in this region is often clinical. The clinical findings of common pseudoaneurysm are a pulsatile mass and an audible thrill, but the location of the artery makes it hard to diagnose, so it is confirmed by radiological studies such as computed tomography and angiography. In both cases, angiography allowed the exact position of the defect to be identified and selectively embolized, maximally preserving normal vasculature. The angiogram also excluded other vascular anomalies or malformations that may have complicated the patients' management [1,3,5,6,12,14].

The initial treatment for these patients is not different from any other traumatic patient, such as airway, ventilation, and management of shock, which includes hemorrhage control. Hemorrhage control in the form of endoscopic arterial embolization is an emerging popular treatment. Embolization should be performed as close to the bleeding point as possible



Fig. 3 – Preoperative computed tomography scan of the facial bones in a 24-year-old male patient presenting with nasal fracture, bilateral zygomatic fracture, and right mandibular parasymphisis fracture following a blunt trauma.



Fig. 4 – The angiography demonstrated another extravasation from a pseudoaneurysm of the left sphenopalatine artery (arrow).

to avoid complications such as cranial nerve palsies. Additionally, superselective distal embolization may not stop the bleeding more rapidly than proximal embolization, leaving the risk of recurrent bleeding from anastomotic channels distal to the embolization site. Treatment is also crucial in preventing permanent injury such as visual disturbance or vision loss. Like any other injury that requires hemorrhage control, transarterial or transvenous embolization using coils, acrylic glue, or Onyx has proven useful in relieving the damage of CCF. This study took a careful approach to diagnosing the condition to prevent life-threatening or seriously damaging complications to the eyes. Embolization has also proven effective in managing patients' complaints [15]. Overall, we aimed to provide insight for those who may be facing similar challenges, with the latest treatment offering maximal therapeutic benefit.

Conclusion

Massive recurrent epistaxis, which can manifest after a head injury, can be caused by pseudoaneurysm rupture. The diagnosis of pseudoaneurysm is clinical; thus, confirmation by radiological studies such as angiography prompts urgent management. Endovascular embolization, either transarterial or transvenous, is believed to be an effective treatment for massive recurrent epistaxis. It can also prevent the progression of any life-threatening complications.

Patient consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patients.

REFERENCES

- Kusuma Dewi AM. Epistaksis masif pada pseudoaneurisma traumatik arteri karotis interna. Oto Rhino Laryngol Indones 2016;46:184–91. doi:10.32637/orli.v46i2.166.
- [2] Tunkel DE, Anne S, Payne SC, Ishman SL, Rosenfeld RM, Abramson PJ, et al. Clinical practice guideline: nosebleed (epistaxis). Otolaryngol Neck Surg 2020;162:S1–38. doi:10.1177/0194599819890327.
- [3] Reyre A, Michel J, Santini L, Dessi P, Vidal V, Bartoli J-M, et al. Epistaxis: the role of arterial embolization. Diagn Interv Imaging 2015;96:757–73. doi:10.1016/j.diii.2015.06.006.
- [4] Nicolay S, Van Der Zijden T, Voormolen M, D'Archambeau O, Maes J, De Belder F, et al. The endovascular treatment of epistaxis. J Belgian Soc Radiol 2015;99:65–73. doi:10.5334/jbr-btr.954.
- [5] Chun JJ, Choi CY, Wee SY, Song WJ, Jeong HG. Embolization for treating posttraumatic pseudoaneurysm of the sphenopalatine artery. Arch Craniofac Surg 2019;20:251–4. doi:10.7181/acfs.2019.00227.
- [6] Souza RRL de, Pinto PS, Viana R de S, Brito FRC, Lessa TCS, Dias OAA, et al. Pseudoaneurisma de artéria esfenopalatina após agressão por arma de fogo em face. Res Soc Dev 2020;9:e4959116939. doi:10.33448/rsd-v9i11.6939.
- [7] Gangaraj SS, Pillai N. Intractable epistaxis secondary to traumatic pseudoaneurysm of internal carotid artery. An Int J Clin Rhinol 2021;12:63–5. doi:10.5005/jp-journals-10013-1362.

- [8] Zhang Cw, Xie Xd, You C, Mao By, Wang Ch, He M, et al. Endovascular treatment of traumatic pseudoaneurysm presenting as intractable epistaxis. Korean J Radiol 2010;11:603–11. doi:10.3348/kjr.2010.11.6.603.
- [9] Agha RA, Franchi T, Sohrabi C, Mathew G, Kerwan A, Thoma A, et al. The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. Int J Surg 2020;84:226–30. doi:10.1016/j.ijsu.2020.10.034.
- [10] Oley MC, Oley MH, Durry MF, Kepel REM, Faruk M. Cutaneous angiosarcoma: a case report. Int J Surg Case Rep 2021;88:106506. doi:10.1016/j.ijscr.2021.106506.
- [11] Swain SK, Das A, Acharya S, Shajahan N, Agrawala R. Acute onset of massive epistaxis due to post-traumatic cavernous internal carotid artery pseudoaneurysm: a case report. J Acute Dis 2021;10:39. doi:10.4103/2221-6189.307393.
- [12] Abrich V, Brozek A, Boyle TR, Chyou P-H, Yale SH. Risk factors for recurrent spontaneous epistaxis. Mayo Clin Proc 2014;89:1636–43. doi:10.1016/j.mayocp.2014.09.009.
- [13] Kim Y-W, Baek M-J, Kim H-D, Cho K-S. Massive epistaxis due to pseudoaneurysm of the sphenopalatine artery: a rare post-operative complication of orthognathic surgery. J Laryngol Otol 2013;127:610–13. doi:10.1017/S0022215113000819.
- [14] Goh S, Ritchie L, Joseph T, Brookes J. Spontaneous sphenopalatine pseudoaneurysm: recurrent epistaxis in a patient with cryptogenic vasculopathy. BMJ Case Rep 2015:bcr2015210159. doi:10.1136/bcr-2015-210159.
- [15] Belhachmi A, Dokponou Y, Imoumby F, Imbunhe N, Akroud S, Gazzaz M. Severe traumatic brain injury with sphenopalatine artery ruptured: case report and review of the literature. Open J Mod Neurosurg 2021;11:204–9. doi:10.4236/ojmn.2021.113024.