International Journal of Emergency Medicine

RESEARCH Open Access

Traumatic facial arteriovenous fistula: a rare case report and literature review

Hongwei Gao[1,3], Donglin Zhou[2], You Zhou[1,3], Xuechao Wu[1,3], Jing Wang[1,3] and Qing Wang[1,3,4*]

Abstract

The formation of facial arteriovenous fistula (AVF) directly between the facial artery and vein after trauma is very rare. Compared with intracranial AVF, understanding of this condition is limited. This paper reported the case of a 49-year-old male who had a metallic foreign object impaled on his left neck while weeding 6 months prior to admission. However, 3 months prior to admission, he developed a gradual throbbing of the left neck and swelling on the left side of his face. Auscultation revealed a vascular murmur in front of the mandibular angle on the left side of the face. Digital subtraction angiography (DSA) showed an AVF between the left lateral facial artery and vein, resulting in retrograde blood flow into cavernous sinus though the ophthalmic vein. The authors discuss the management of a traumatic AVF through combined transarterial embolization using coils and Onyx liquid embolic agent. A 3-month follow-up indicated no recurrence of AVF, and the patient had a great recovery with normal-appearing left face and eye. It was the isolated involvement of the facial artery and vein in a post-traumatic setting makes this case particularly instructive. In addition, we summarized the clinical symptoms and treatment of AVF in the face and neck regions.

Keywords Facial arteries, Facial vein, Arteriovenous fistula (AVF), Embolization

Introduction

Arteriovenous fistula (AVF) is a disease caused by abnormal connection between arteries and veins [1]. In this disease, blood flows directly from the high-flow arterial system into the low-flow venous system, bypassing the capillary system [2]. This leads to symptoms such as reduced arterial blood supply and obstruction of venous return. The major causes of AVF may be trauma and iatrogenic injury [3]; however, it can occur spontaneously as well. In fact, compared with intracranial AVFs, extracranial AVFs generally have definite injury factors, including puncture wound and blunt trauma, and low incidence rate [2, 4, 5]. Even if AVFs are formed due to some external causes, the structure of feeding arteries and draining veins may be complicated because the tissues of head and neck are relatively vascularized. However, AVFs consisting of a single artery and vein are rare after trauma in the region of head and neck, except for the case described in this report. Sim SY et al. [6] pointed out that the incidence rate of intracranial dural arteriovenous fistula was dominated by transverse sinus (50%), cavernous sinus (16%), tentorium cerebellum (12%) and superior sagittal sinus (8%). However, there is currently no definitive authoritative overall data on the incidence of facial arteriovenous fistula (AVF), which not only reflects the relatively rare occurrence of facial AVF in

*Correspondence:
Qing Wang
wxwqnj@163.com
[1]Wuxi School of Medicine, Jiangnan University, Wuxi, Jiangsu 214002, China
[2]Department of Neurosurgery, Qilu Hospital, Cheeloo College of Medicine, Institute of Brain and Brain-Inspired Science, Shandong University, 107 Wenhua Western Road, Jinan, Shandong 250012, China
[3]Wuxi Neurosurgical Institute, Wuxi, Jiangsu 214002, China
[4]Department of Neurosurgery, Jiangnan University Medical Center, The Wuxi No.2 People Hospital, Wuxi, Jiangsu 214002, China

actual clinical diagnosis and treatment, but also implies that due to the small number of cases, the accumulated diagnostic and treatment experience of medical staff is relatively insufficient. Reporting such rare cases has important clinical and scientific value. Although the cases of complex facial AVF have been treated with both intravascular embolization and surgical resection [2, 7], it is still necessary to design individual treatment plans based on the extracranial area and complexity of the AVF. In this case report, AVF was successfully embolized using coils combined with Onyx glue. In addition, we reviewed various treatment options for AVF in the maxillofacial region to enhance understanding of this disease.

Case report

A 49-year-old male complained of throbbing tremors on the left side of his face for 6 months and increased facial swelling for 3 months after the trauma. The patient experienced sharp pain on the left side of his neck while weeding 6 months prior to admission. The wound was pointed and bleeding profusely. At that time, the patient pressed the wound to stop the bleeding, but no other treatment was received. However, he subsequently developed pulsations in the left cheek and mandible region, which were consistent with heartbeat. Further, 3 months prior to admission, the patient had significant swelling on the left side of the neck and face, accompanied by red conjunctiva in the left eye and swelling and bluish upper lip. Physical examination revealed swelling in the left conjunctiva and vascular murmurs in the mandibular region; however, free movement of the eye in each direction and no neurological dysfunction were reported (Fig. 1). Computed tomography (CT) revealed a surprising presence of a metallic foreign (A hard iron foreign object resembling a cylinder, with a maximum diameter of approximately 60 × 25 mm.) body on the left side of the neck. Furthermore, CT angiography (CTA) of the head and neck was performed to identify intracranial secondary lesions and to determine the position of metallic foreign bodies in the neck vessels. Fortunately, the metallic foreign body appeared to have been punctured from the carotid triangle in the direction of the mastoid process; however, no evidence of damage to the large blood vessels in the neck was observed, significantly reducing the risk of surgical removal. Whole-brain digital subtraction angiography (DSA) was performed to obtain a definite clinical diagnosis.

The vascular composition of traumatic AVF was studied in detail using DSA. The traumatic fistula was established between the left external carotid branch facial artery and facial vein. The blood flowed directly and rapidly into the facial vein, and an enlarged venous lake was observed at the beginning of the reflux vein. The facial vein was clearly curved and dilated and drained to the intracranial veins through the ipsilateral ophthalmic vein. At the same time, superficial veins, such as the contralateral lateral vein, drained into the contralateral external jugular vein. The DSA images are given in Fig. 2. the blood is drained through other facial superficial veins into the lateral external jugular vein.

First, the stereotactic technique of the robotic system was used to accurately localize the foreign body in the neck (Fig. 3). An 8-cm incision was designed from the mandible to the mastoid process. Further, the sternocleidomastoid muscle was exposed to allow easy detection of foreign bodies in the direction of the mastoid process. The surrounding adhering tissue was carefully dissociated until the foreign body was completely removed. Rust and yellow stains were observed on the surface of the foreign body, indicating possible infection. The pus was subjected to bacterial culture and drug sensitivity testing. The operating cavity was rinsed alternately with hydrogen peroxide and iodophor, and the subcutaneous drainage tube was drained and sutured. The second procedure was interventional therapy. We introduced two Echelon-10 microcatheters into the proximal and distal parts of the fistula via the femoral artery using a Traxcess 0.014 micro-guidewire. Through the proximal echelon-10 microcatheter, three coil springs (Hydro Frames 6 mm × 23 cm, 5 mm × 15 cm, and 4 mm × 12 cm) were released to fill the AVF and reduce blood flow. However, after 1.5 mL Onyx glue was injected through the proximal catheter, angiography revealed that the distal orifice fistula was still developing (Fig. 4). Therefore, the distal echelon-10 microcatheter was injected with 1 mL Onyx glue for embolization and reinforcement of the partial facial vein. Fortunately, imagingIntraoperative angiography confirmed that the fistula had completely disappeared, and the tortuous and dilated facial veins had not developed again. The treatment process is given in Figure. Postoperative treatment included tetanus antitoxin and antibiotics. One day after the surgery, the patient's facial pulsation was significantly diminished without neurologic deficits, and the embolized coils and glue could be reached under the left jaw. Hyperemia in the left conjunctiva and facial swelling on the left side improved 3 months after the surgery.

Discussion

Traumatic AVF in the maxillofacial region is fairly rare and is commonly attributed to different degrees of trauma in past, including penetrating wounds [1, 2, 8], blunt trauma [4, 5, 9], or even iatrogenic injury [3, 4, 10–12]. These injury factors may immediately induce or indirectly promote the construction of abnormal vascular pathways in the maxillofacial region. The clinical symptoms of traumatic AVF are mainly shunt of the arterial system and disturbance of the circulation in the venous

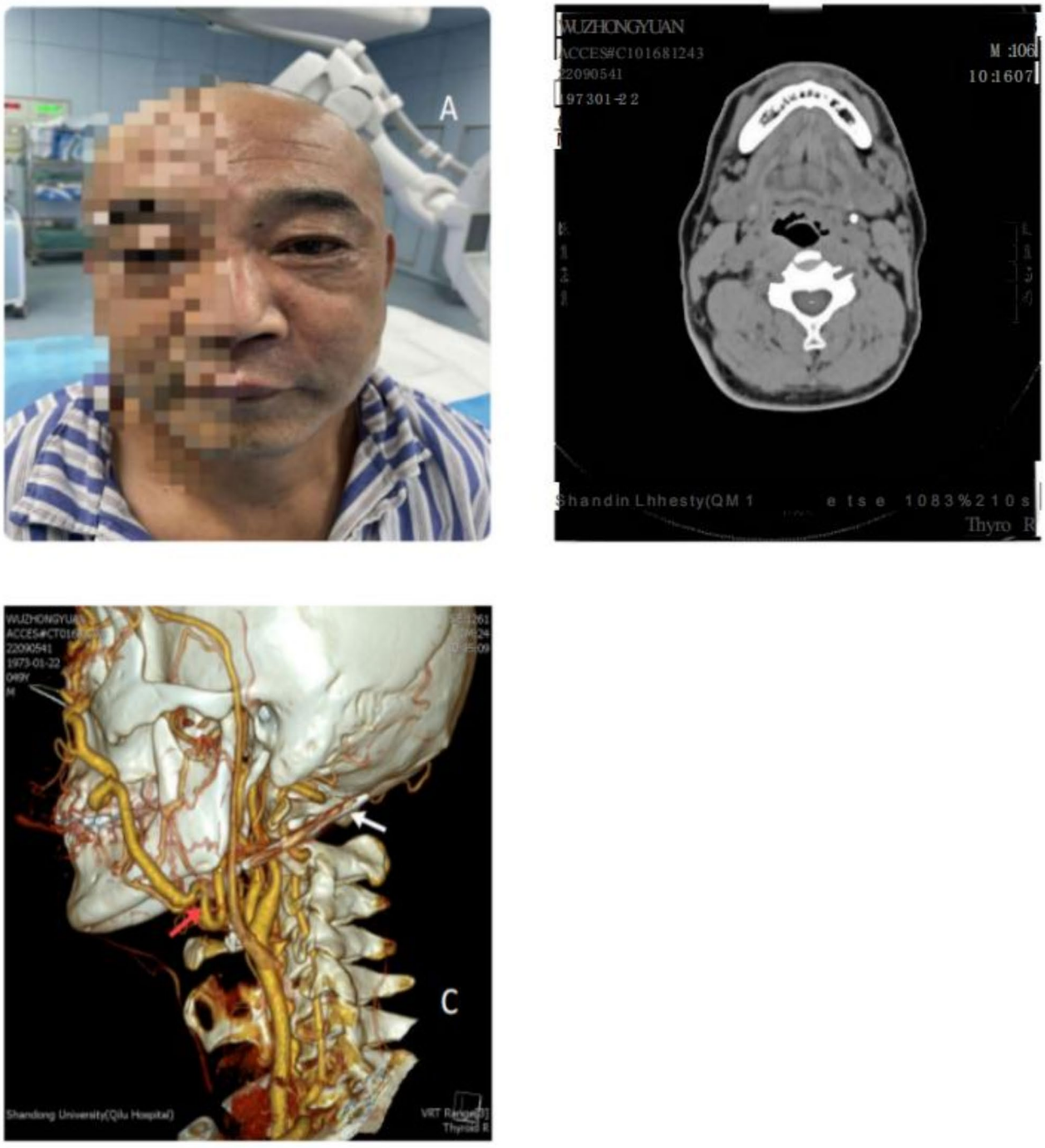

Fig. 1 Preoperative facial image and head computed tomography (CT) images. (**A**) The patient's facial image indicated redness and swelling on the left side of the face with conjunctival congestion in the left eye. (**B**) Axial section's projection of the neck and (**C**) three-dimensional reconstruction of neck vessels. The red arrow indicates the fistula orifice and the white arrow indicates a metal foreign body, which punctures facial arteries and veins and heades toward the mastoid process without injuring the neck vessels

system; they depend on the degree and location of injury and complexity of fistula. Clear etiologies and early clinical symptoms are the characteristics of this disease; therefore, early intervention is possible and necessary.

Usually, for simple and small facial AVFs, the complete occlusion rate can reach relatively high levels, possibly 70 – 90% or even higher in some studies. But for complex, large, or those with multiple feeding arteries, the complete occlusion rate may be around 40 – 60%. The cure

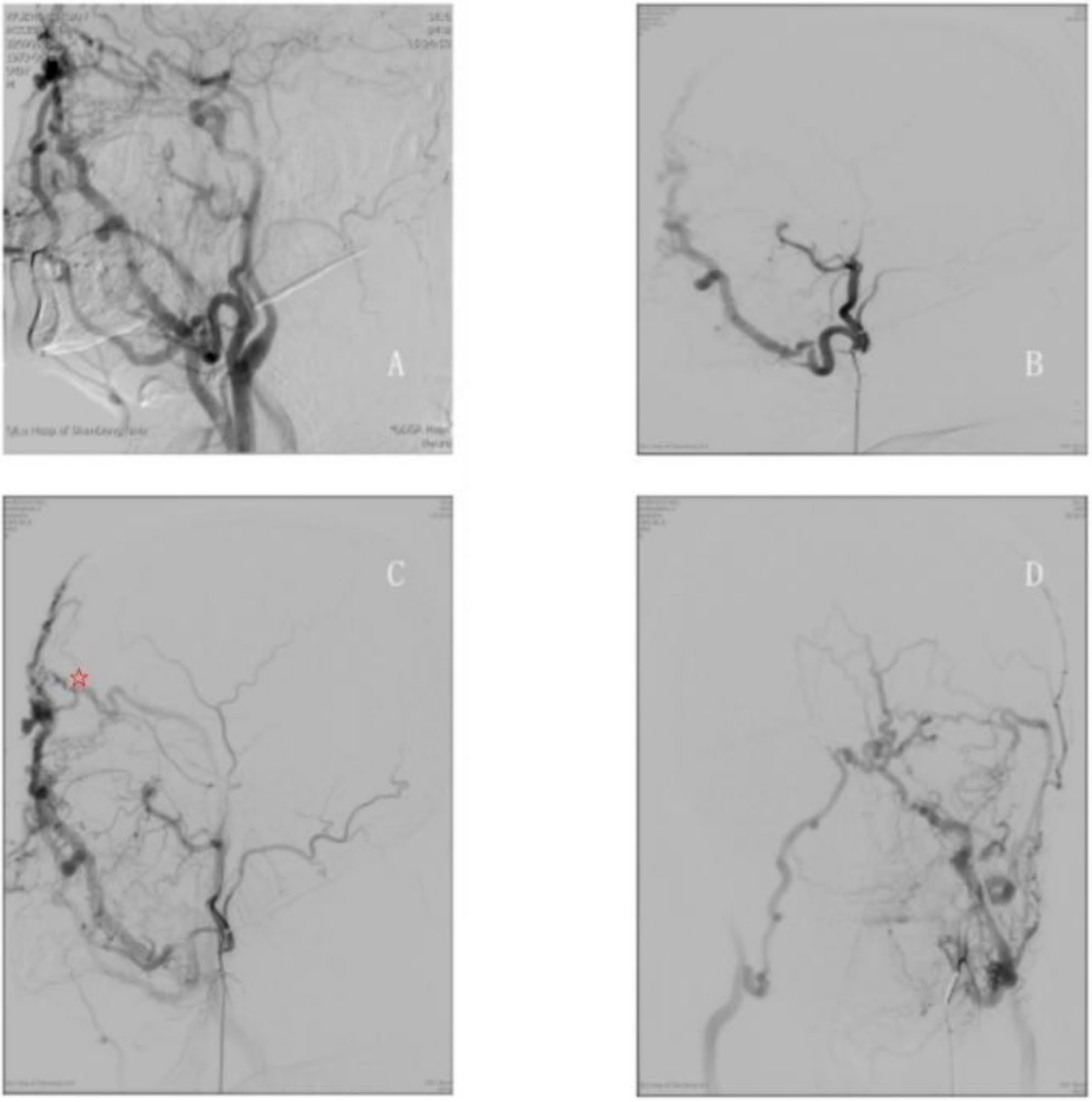

Fig. 2 Preoperative DSA images. (**A**) Lateral external carotid artery angiography indicates AVF, supplied by the left facial artery. (**B**) In the venous phase, the left projection of the face indicates blood flow to the facial vein, making it tortuous and dilated. (**C**) The blood then flows back into the left ophthalmic vein and into the intracranial vein. The star indicates the ophthalmic vein. (**D**) The anterior projection of the face indicates that the blood drains through the superficial facial veins to the right lateral veins and eventually into the right external jugular

rate for percutaneous embolization treatment of congenital facial arteriovenous fistulas exceeds 95% [13]. The choice of embolization materials and techniques matters. For instance, the use of liquid embolic agents like Onyx may achieve better occlusion effects than traditional particulate materials. The complications of AVF may include the following points: Firstly, there may be local tissue ischemia and necrosis. In addation, Embolization agents may cause vascular occlusion in non-target vessels, leading to vascular embolism. What's more facial nerve injury is possible.

With regard to the mechanism of formation of post-traumatic AVF in preauricular region, Takuy and Naoya proposed two potential mechanisms for the development of traumatic AVF [5]. First mechanism is that foreign bodies damage arteries and veins at the same time, and

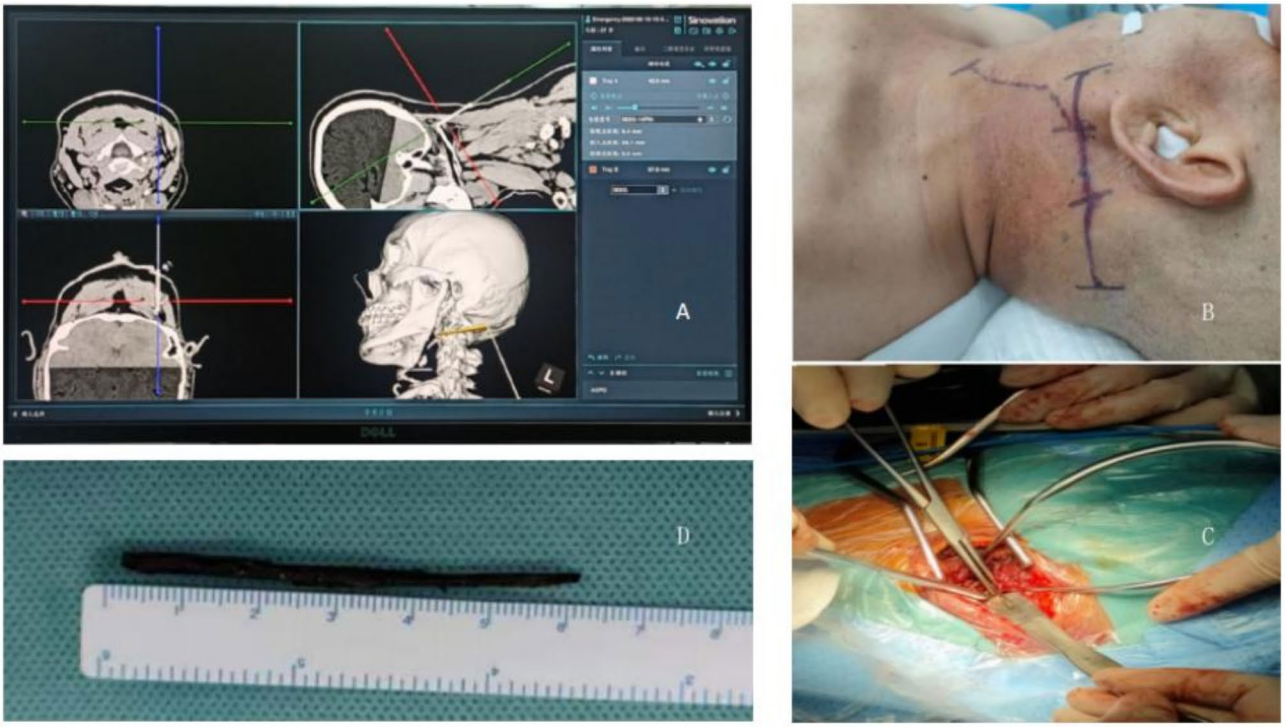

Fig. 3 Photograms in the first operation therapeutic procedure. (**A**) Photogram reveals The image shows the application of robotic positioning system during surgery, which can accurately locate the location of foreign bodies. (**B**) Based on the location of the incision designed by the localization system, the photogram reveals that the orientation is consistent with the foreign body. (**C**) Intraoperative photograph of removal of metallic foreign body from neck. (**D**) Rust and pus on the surface of the foreign body

these vessels anastomose incorrectly to form a fistula. The other mechanism is that the suture used to close the wound penetrates both arteries and veins, leading to fistulas. In addition, Ashish and James E. the suboptimal management of acute injurythe claim clam of the suboptimal management of acute injury coincides with the above two points [3]. In combination with the above views, we speculated that in our case, the metal foreign body might have punctured both the facial vein and artery at the same time and compressed the hemostasis to promote the anastomosis of the arteriovenous puncture sites. The histopathology of AVFs reveals that damaged arterial endothelial cells proliferating and migrating to the peripheral hematoma form endothelial buds and small vessels to establish vascular channels with adjacent veins [4, 10, 14]. With the arteriovenous shunt, the continuous development of the fistula leads to the expansion and thickening of the drainage vein that tends to arterialization [1, 3, 10]. Due to the lack of venous flaps [15, 16], the high flow rate of facial artery constantly impinges on the facial venous system, resulting in increased pressure on the facial vein and its expansion and tortuosity. Obstructed venous return exacerbates facial swelling and numbness. Although blood can be expelled through the ipsilateral external jugular vein, due to the large volume of arterial blood flow, venous DSA indicates that the facial vein drains into the intracranial region via the ipsilateral ocular vein. This results in slightly increased intracranial venous sinus pressure and manifestations such as swelling of the eyelids and conjunctival congestion. Additionally, AVF drains through superficial facial veins to the lateral external jugular vein. Therefore, vascular injury, local hemodynamic changes, and special anatomical structures adjacent to facial arteries and veins contribute to the occurrence and development of this reported traumatic AVF.

DSA remains the gold standard for identifying feeding arteries and draining veins in traumatic AVF. Before angiography primary causes of traumatic AVF, such as foreign bodies and surgical sutures, should be carefully examined and removed, which may aggravate the development of AVF. It is important to not only understand the proximal supply arteries of the AVF but also perform selective angiogram of any other potential distal collateral vessels during angiography [3]. Potential collateral vessels, particularly for maxillofacial traumatic AVF, may originate in the branches of the ipsilateral internal and external carotid arteries, with the lateral vessels involved as well. In addition, there may be other vascular diseases caused by hemodynamic changes around AVFs, such as aneurysms and vascular occlusion [2]. Some injuries to intracranial vessels in trauma cannot be ignored; for example,

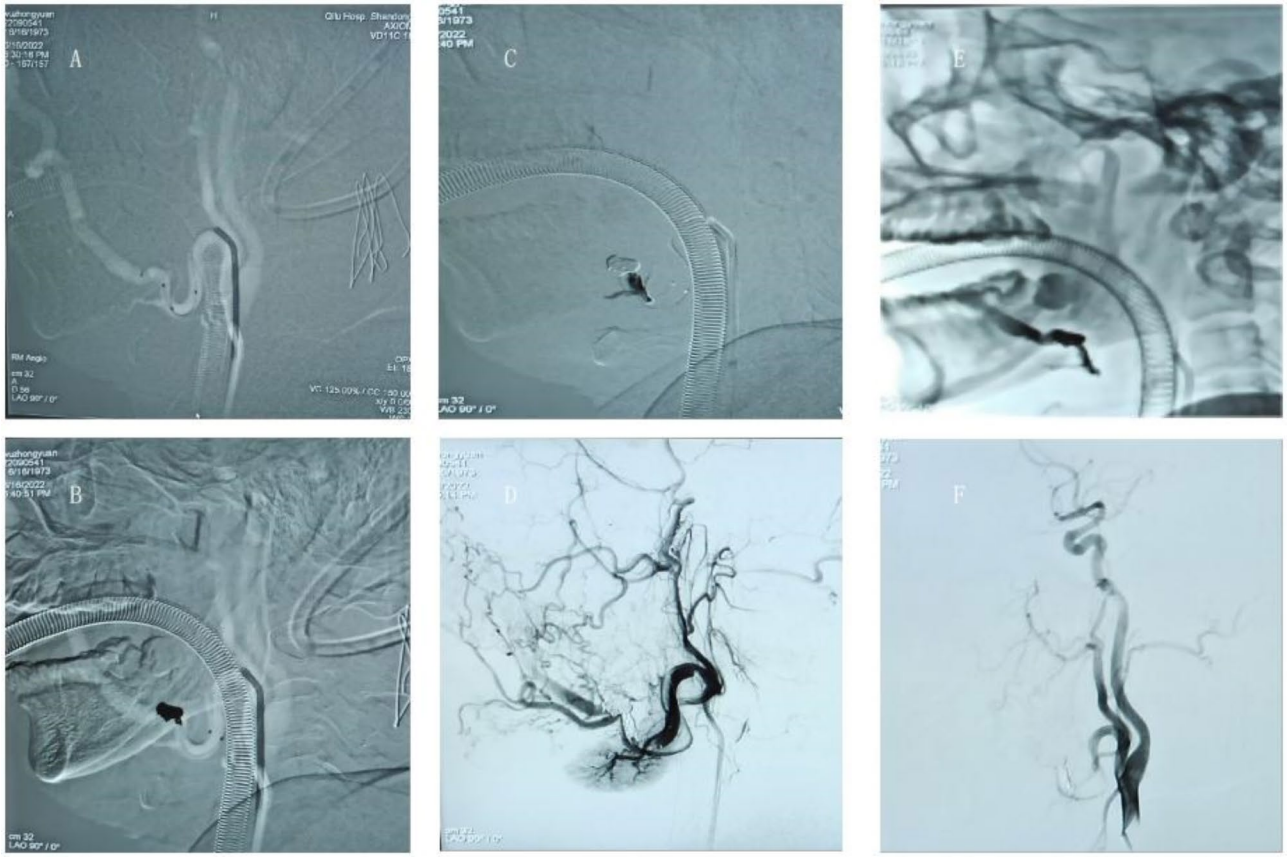

Fig. 4 Angiograms in the second endovascular treatment. (**A**, **B**) The angiograms indicate the two microcatheters reaching the proximal and distal ends of the fistula guided by the microguide wire, and releasing the three coils. (**C**, **D**) Angiography after proximal microcatheter injection of Onyx indicated that the fistula was not completely occluded. (**E**, **F**) The angiograms reveal the distal injection of Onyx until there is no abnormal flow through the fistula orifice

early clinical symptoms of carotid-cavernous fistula (CCF) resemble those of partial maxillofacial AVF.

The principle of arteriovenous therapy in trauma is to control the primary cause, eliminate symptoms, and reduce blood shunting. Traditional surgery and endovascular therapy have been effective treatments for these diseases. The former procedure involves ligation of the vessel and resection of the fistula [7, 17], which may cause venous injury. However, the latter involves embolization of the fistula mainly using coils through the transarterial access [4]. Transvenous access is a suitable alternative when trauma results in occlusion of the proximal artery or when accessing the artery is very difficult. However, for complex traumatic AVFs with excessive blood flow, combination therapy can be effective in reducing the recurrence rate [3, 5, 7]. In a previous study [5], only the proximal superficial temporal artery was embolized at the patient's first visit. Other distal collateral vessels form a vascular network around the fistula, gradually enhancing the blood supply to the AVF; thus, that patient's symptoms persisted. In our case, we removed the fistula using a combination of operations and the significant improvement in the patient's symptoms, the restoration of normal blood flow dynamics as evidenced by imaging studies. In the end achieved excellent therapeutic results. When the instability of the hemodynamics causes difficulty in embolization, the cases of preauricular complex AVFs have been reported that were treated with embolization after collateral vessel ligation to reduce blood flow [2]. The peculiarity of this case lies in the fact that, in contrast to surgical injuries and open trauma, the foreign body caused only facial artery and vein tears with compressive hemostasis, all of which led to poor anastomosis. Consequently, the treatment of single AVF highlights the need for complete embolization. Given the complex anatomical relationship between facial nerves and vessels and infection of metal foreign body, ligation of the fistula was not performed when the foreign body was removed. We chose to embolize the fistula with a spring coil to first reduce blood flow and then inject Onyx glue into the proximal and distal vessels of the fistula to fully seal it. The embolization with coil and Onyx glue has been widely used to treat large aneurysms, arteriovenous malformations, and CCF. The possibility of migration

after Onyx glue injection and incomplete embolization of coils must be considered while treating large fistulas with unstable blood flow. In our case, the AVF was located in the extracranial region; therefore, the treatment was relatively safe without the space-occupying effects caused by coil and Onyx glue insertions. Moreover, for this case, postoperative prevention of facial nerve injury and maxillofacial soft tissue infection is the direction of focus during long-term follow-up.

Conclusion

Traumatic facial AVFs in the maxillofacial region are rare and are mostly caused by foreign body injuries and iatrogenic injuries. Comprehensive angiography and individualized treatment assure reduced recurrence rates. The combination of coils and Onyx glue may be an appropriate treatment for AVFs with excessive blood flow. Detailed case reports provide reference for the development of treatment plans, especially in the absence of large-scale clinical trial data, where these case experiences may become important decision-making basis. Secondly, the accumulation of rare cases helps us better understand the pathogenesis, natural course of the disease, and points out the direction for future research.

Abbreviations

AVF	Arteriovenous fistula
CT	Computed tomography
CTA	Computed tomography angiography
DSA	Digital subtraction angiography
CCF	Carotid-cavernous fistula

Acknowledgements

Not applicable.

Author contributions

All authors contributed to data collection. The article was written by HG with assistance from DZ, taking into account the comments and suggestions of the coauthors. QW, YZ, XW and JW prepared Figs. 1, 2, 3 and 4. All coauthors had the opportunity to comment on the analysis and interpretation of the findings and approved the final version for publication.

Funding

No Funding.

Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

The ethical approval for reporting this case comes from the Ethics Committee on Scientific Research of Qilu Hospital of Shandong University (No. KYLL-2021(ZM)-293). Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal.

Consent for publication

All presentations of case reports have consent for publication.

Competing interests

The authors declare no competing interests.

Received: 4 December 2024 / Accepted: 11 March 2025
Published online: 01 April 2025

References

1. Wang C, Ge Y, Li L. Traumatic arteriovenous fistula of the left Parotid region: A case report[J]. Oncol Lett. 2016;11(5):3194–6.
2. Guo J, Zhan R, Ren Y, et al. The hybrid technical management of large and complicated traumatic arteriovenous fistula of preauricular Region[J]. J Craniofac Surg. 2018;29(2):432–6.
3. Saini A, Jackson JE. Arteriovenous fistulas of the facial artery after mandibular surgery: treatment by embolization[J]. AJR Am J Roentgenol. 2008;190(1):W35–40.
4. Schwartz GF, Rankow RM. Traumatic arteriovenous fistula of the facial artery[J]. Plast Reconstr Surg. 1967;40(5):453–6.
5. Akai T, Kuwayama N, Endo S, et al. Treatment of traumatic facial arteriovenous fistula with embolization and surgical excision–a case report[J]. J Craniomaxillofac Surg. 1994;22(3):147–50.
6. Sim SY, Cerebrovasc Endovasc J. Neurosurg. 2022;24(3):203–9. https://doi.org/10.7461/jcen.2022.E
7. Robbs JV, Carrim AA, Kadwa AM, et al. Traumatic arteriovenous fistula: experience with 202 patients[J]. Br J Surg. 1994;81(9):1296–9.
8. Whiteside OJ, Monksfield P, Steventon NB, et al. Endovascular embolization of a traumatic arteriovenous fistula of the superficial Temporal artery[J]. J Laryngol Otol. 2005;119(4):322–4.
9. Amirjamshidi A, Abbassioun K, Rahmat H. Traumatic aneurysms and arteriovenous fistulas of the extracranial vessels in war injuries[J]. Surg Neurol. 2000;53(2):136–45.
10. Janssen M, Vaninbroukx J, Fourneau I. Arteriovenous fistula after superficial Temporal artery biopsy[J]. Ann Vasc Surg. 2013;27(4):500–1.
11. Moses JJ, Topper DC. Arteriovenous fistula: an unusual complication associated with arthroscopic temporomandibular joint surgery[J]. J Oral Maxillofac Surg. 1990;48(11):1220–2.
12. Preisler SA, Koorbusch GF, Olson RA. An acquired arteriovenous fistula secondary to temporomandibular joint arthroscopy: report of a case[J]. J Oral Maxillofac Surg. 1991;49(2):187–90.
13. Braun J, Zuckerman-Levin N, Even-Sapir E, Lorber A. [Congenital arteriovenous facial fistula cured by percutaneous embolization]. Harefuah. 1992;122(7):417-9, 480. Hebrew. PMID: 1592296.
14. Marks MW, Argenta LC, Dingman RO. Traumatic arteriovenous malformation of the external carotid arterial system[J]. Head Neck Surg. 1984;6(6):1054–8.
15. Marur T, Tuna Y, Demirci S. Facial anatomy[J]. Clin Dermatol. 2014;32(1):14–23.
16. Junn JC, Som PM. Maxillofacial skeleton and facial Anatomy[J]. Neuroimaging Clin N Am. 2022;32(4):735–48.
17. Holt GR, Holt JE, Cortez EA, et al. Traumatic facial arteriovenous malformations[J]. Laryngoscope. 1980;90(12):2011–20.

Publisher's note