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The Hybrid Technical Management of Large and Complicated Traumatic Arteriovenous Fistula of Preauricular Region

Jian Guo, MD, PhD, Rucai Zhan, MD, PhD,* Yong Ren, MD,† Xiangjing Meng, MD,* Rengli Liu, MD, PhD,* Guijie Li, MD,‡ Dong Wang, MD,§ and Jinlong Sun, MD, PhD**

Background: Arteriovenous fistula (AVF) is defined as an abnormal communication between the high flow arterial system and the low flow venous network, which directly connects the arterial feeding vessels and the near draining veins without normal intervening capillary bed. Arteriovenous fistula incurs in preauricular region is exceeding rare. Most of these fistulae occur as a result of an iatrogenic injury, the volume is small, feeding and draining vessels of feeding and draining are simple, and can be cured easily. However, the treatment of the large and complicated AVF after incidental trauma in preauricular region is a challenge even for senior neurosurgeon. In this study, the authors discuss the management of a traumatic AVF through combined therapeutic method of surgical ligation and transarterial embolization. It is fed by ipsilateral superficial temporal artery, internal maxillary artery, posterior auricular artery, and their accessory branches and is drained by ipsilateral common facial vein and external jugular vein. Also the etiology, clinical manifestations, pathology, diagnosis, and management are summarized.

Conclusion: Large and complicated traumatic AVF in preauricular region is rare, often due from an injury in maxillofacial region, combined therapy needed.

Key Words: Arteriovenous fistula, maxillofacial region, preauricular region, superficial temporal artery, superficial temporal vein

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Arteriovenous fistula (AVF) is defined as an abnormal communication between the high flow arterial system and the low flow venous network, which directly connects the arterial feeding vessels and the near draining veins without normal intervening capillary bed.[1,2] Arteriovenous fistula incurs in preauricular region is exceeding rare. Most of these fistulae occur as a result of an iatrogenic injury,[3–9] the volume is small, feeding and draining vessels of feeding and draining are simple, and can be cured easily. However, the treatment of the large and complicated AVF after incidental trauma in preauricular region is a challenge even for senior neurosurgeon. In this study, we discuss the management of a traumatic AVF through combined therapeutic method of surgical ligation and transarterial embolization. It is fed by ipsilateral superficial temporal artery (STA), internal maxillary artery, posterior auricular artery, and their accessory branches and is drained by ipsilateral common facial vein and external jugular vein. Also the etiology, clinical manifestations, pathology, diagnosis, and management are summarized.

CLINICAL MATERIALS

A 47-year-old man complained a gradually augmented "tremulous" swelling in right maxillofacial region for nearly 20 years, and felt regional throbbing and pulsatile tinnitus in right ear for nearly 15 years. The patient was accidentally pricked in right aural region when playing the game of throwing "flying cutter" which was made of ferric slice with his juvenile partners. On physical examination, there was an approximately 13×8 cm nontender, soft, warm, and pulsatile swelling with a palpable thrill and a continuous machinery-like murmur in synchrony with the heartbeat in right maxillofacial (Fig. 1). It is upper border reaches zygomatic arch, anterior approaches cheeks, posterior reaches retromandibular triangle, and bottom is lower than submandibular triangle. The apogee of the puffiness lies at angle of jaw, while the tremor and murmurs above the neck of condyle are extremely conspicuous and strong. Moreover, the thrill and bruit would disappear when enough pressure was applied on the anterior–inferior border of the tragus and the swelling would also gradually shrink in a few seconds, the varicose external jugular vein would subsequently disappear. A scar, approximately 0.5 cm in length, was present beneath the tragus. A detailed neurologic examination showed no abnormalities. Ultrasonography revealed internal diameter of the thickened and tortuous right external carotid artery (ECA). Thoracic photograph of x-rays revealed normal (Fig. 2). Cranial magnetic resonance imaging (MRI) revealed multiple enlarged vessels in the right maxillofacial region, infratemporal fossa, and some manifold deformans blood flow signals around the right neck of condyle (Fig. 3). There is an aqueous sac with a cross section of about 6×3.5 cm on the surface of mandible and retromandibular triangle region. The right common carotid artery (CCA), external carotid arterial system are all markedly enlarged, and there is a generous popcorn-shaped cavum connecting with the main trunk of STA next to its root. The following digital subtraction angiography found as follow (Fig. 4): the right CCA, ECA vascular system turned remarkably thickened

From the *Department of Neurosurgery; †Department of Dermatology; ‡Department of Interventional Diagnosis and Treatment; and §Department of Cardiac Surgery, Shandong Provincial Qianfoshan Hospital Affiliated to Shandong University, Jinan, China.

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Address correspondence and reprint requests to Jinlong Sun, MD, PhD, Shandong Provincial Qianfoshan Hospital, Jinan, China; E-mail: jinlongsun@hotmail.com

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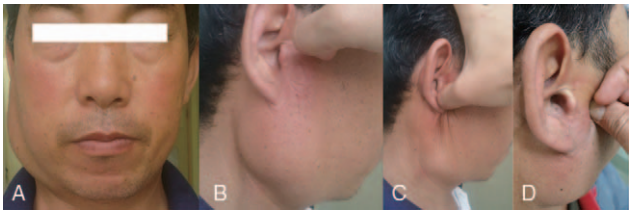

FIGURE 1. Photographs of the lesion projection with (A) frontal and (B-D) lateral shots demonstrating its position, size, change under compression, and the scar. (A) The right maxillofacial region is larger than the left. (B) The circumscription of the lesion. (C) Under enough compression only a small pulsating swelling is residual, which adjoining the angle of mandible, and the well-marked wrinkle indicating the atrophica adipose layer. (D) The scar (←) is situated anteroinferior to the external auditory canal.

and tortuous. There was a generous and irregular vascular mass located next to the root of STA. The grossus ECA main trunk between the irregular vascular mass and the root of occipital artery twisted as a U-shaped. The right STA, internal maxillary artery, and posterior auricular artery were directly or indirectly supplying blood to the vascular mass. The irregular vascular mass, showed in popcorn shaped in MRI angiogram, drained through ipsilateral common facial vein and external jugular vein, which appeared remarkably thickened and tortuous and were further drained into the internal carotid vein and the subclavian vein, respectively. There was a large venous lake, adjoining to the vascular mass, in the regurgitant veins. Right internal carotid artery would appear under enough pressure on the anterior–inferior border of the tragus and would fleetly vanish without compression. The blood, in the segment of STA main trunk that was located beyond the irregular vascular mass, widdershins and centripetally flowed to the vascular mass. It was mainly from the internal maxillary artery and the posterior auricular artery via anastomosis with the STA, while the distal frontal and parietal branches of STA that beyond the anastomosis became very thin. The blood supply of right cupula scalp was partly fed by leftward ECA. No blood of intracranial vessels and contralateral ECA flow to the fistulous mass was found. On the basis of these findings and case history, we diagnosed the lesion as a

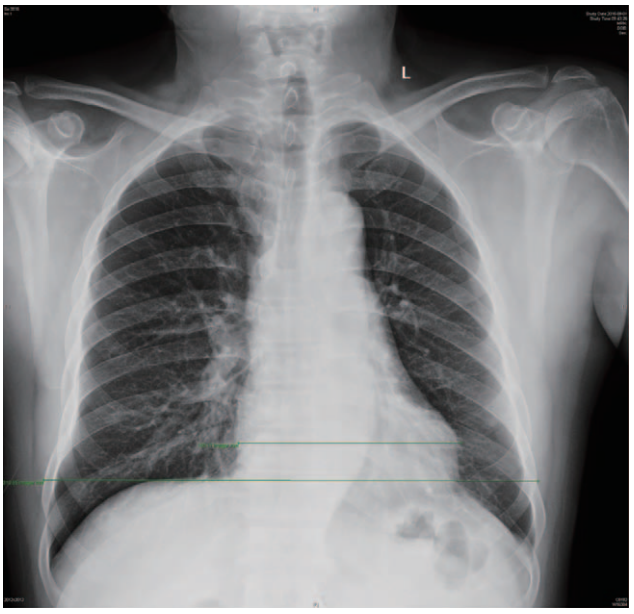

FIGURE 2. Thoracic photograph of x-rays revealed the cardiothoracic ratio was 0.48, which was in the normal physiologic range. And no other cardiopneumatic abnormality was found.

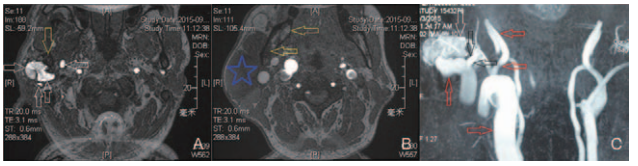

FIGURE 3. (A, B) Axial sections' projections of magnetic resonance imaging and (C) coronal projection of magnetic resonance imaging angiogram. (A) Some manifold grossus and deformans blood flow signals (white arrow) appearing around the right neck of condyle (yellow arrow) and in the infratemporal fossa. (B) There is an aqueous sac (blue ☆) with a cross section of about 6 × 3.5 cm on the surface of mandible (yellow arrow) and retromandibular triangle region. (C) The right common carotid artery (red "→"), external carotid arterial system are all enlarged, and there is a generous popcorn-shaped cavum (white "↓") directly connecting with the main trunk of superficial temporal artery (red "↑") right next to the root of internal maxillary artery (black arrow). The right internal carotid artery (red "←") looks normal.

traumatic AVF in right preauricular region that involving the main arteries and veins in maxillofacial region. The irregular vascular mass, which directly connected the feeding arteries and draining veins, just was the orificium fistulae.

At first, we selected transarterial embolotherapy for the AVF and consumed one 20 mm × 40 cm Interlock-35 coils (Detachable coil-cube; Boston Scientific, Marlborough, MA) and 3 No 2 detachable balloon (Balt, Montmorency, France), but failed (Fig. 5). A month later, we performed a combined therapy of vascular ligation and intravascular embolism for him.

The therapy was also implemented under general anesthesia in Invasive Technology Department. Initially a transfemoral angiography catheter was implanted into the right CCA to monitor the therapeutic process, at first reviewed visualization obviated new change in the lesion. The intraoperative process included 3 procedures (Fig. 6). The first procedure was to dissect and exposure relative blood vessels for immediately ligation. Three incisions were made: the first incision, from rear of the angle of mandible to the intersection point of anterior border of sternocleidomastoid and horizontal line of cricoid cartilage, was for revealing the proximal part of common facial vein and the lowest segment of U-shaped loop, these 2 vessels were all circled cord for ligature. The second incision, along marked STA main trunk above the zygomatic arch, where STA main trunk was dissected and circle with suture at its proximal part. The third incision was made at proximal part of external jugular vein along superior border of clavicle, where we revealed and circled the distal end of external jugular vein with No 10 silk suture. Second procedure was to ligate these vessels. After heparinization through peripheral vein we immediately ligated these vessels in order: STA trunk with No 7 silk suture, ECA doubly with double strands No 10 silk suture. We subsequently crushed and eliminated blood in the dilatated lumen of common facial vein, external jugular vein, and ligated them, respectively, at centripetal distal end.

Third procedure was interventional therapy. We punctured ECA at centrifugal side of ligating silk suture and implanted 6F sheath into the ascending portion of U-shaped loop, just approximating the orificium fistulae, this path was used for embolism and contrast examination. At last, we confirmed the orificium fistulae was soundly embolized, consuming 6 detachable Interlock-35 coils (20 mm × 40 cm × 4, 18 mm × 40 cm × 2 Detachable Coil-Cube, Boston Scientific), through contrast examination. In the procedure the venous lake swollen with tension rising, thrice suctions were given and about 25 mL mixed liquor of blood and contrast media was sucked out. We sewed up these 3 incisions in layered fashion with a standalone drain, which was kept for near 30 hours, in the first incision. The patient awoke from general anesthetic without neurologic deficits, and tinnitus had sheerly vanished.

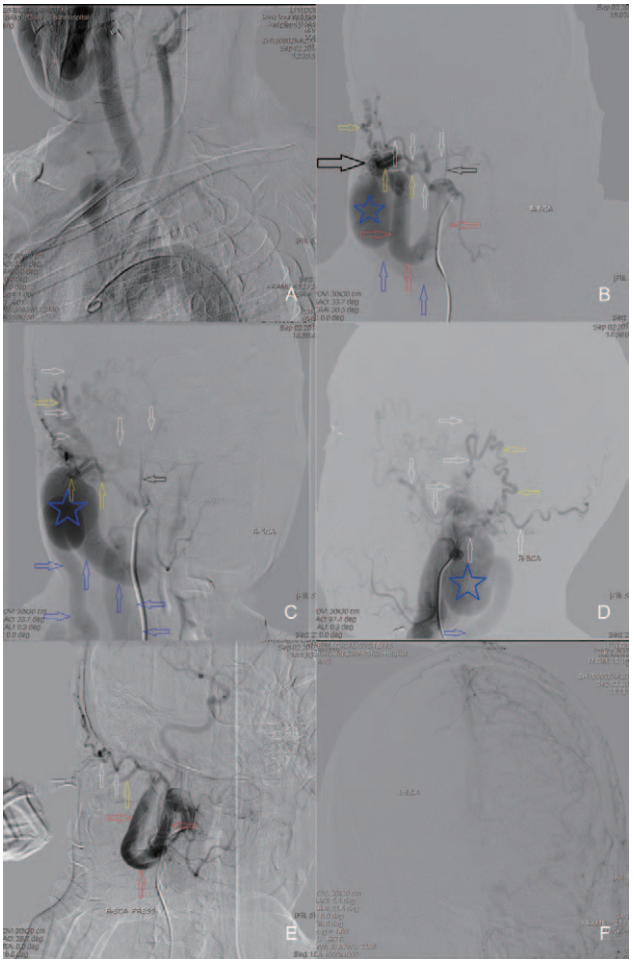

FIGURE 4. The patient underwent a following digital subtraction angiography. (A) The right common carotid artery , external carotid artery vascular system were remarkably thickened and tortuous. (B) There was a generous and irregular vascular mass (black "→"), popcorn-shaped in MRA, directly connected with the STA trunk just next to the root of internal maxillary artery (white "↓"). A grossus U-shaped arterial loop (red arrow) appeared in ECA between the irregular vascular mass and the root of occipital artery (white "↑"). The ascending pharyngeal artery looks normal while the posterior auricular artery looks circuitous and thickened. There was a large venous lake (☆), the aqueous sac in magnetic resonance imaging, appeared in the regurgitant veins that adjoining to the irregular vascular mass. (C) The right STA (white "→") and internal maxillary artery (white "↓") were directly or indirectly supplying blood to the vascular mass and it was drained through ipsilateral common facial vein (blue "↑") and external jugular vein (blue "→"), which appeared remarkably thickened and tortuous, and were further drained into the internal carotid vein (blue "←") and the subclavian vein, respectively. (D) It was through much anastomosis the internal maxillary artery (white"↓") and posterior auricular artery (yellow arrow) connect with the STA (white "→") that beyond the irregular vascular mass and the blood in the STA trunk widdershins and centripetally flowed to the vascular mass, while the distal frontal and parietal branches of STA that beyond the anastomosis became very thin. (E) Right internal carotid artery appeared under enough pressure on the front of the tragus and would fleetly vanish without compression. (F) The blood supply of right copula scalp was partly fed by leftward external carotid artery. There was no blood from intracranial vessels and no contralateral external carotid artery flow to the fistulous mass. STA, superficial temporal artery.

After operation the patient was given strict blood pressure control at 110 to 130/60 to 90 mm Hg, right about 20 mm Hg below preoperative fundamental height; 5000 U low molecular heparin was subcutaneously injected bis in die in first postoperative week and once a day in the following week. His complains of postoperative apparente headache and jumping pain in right

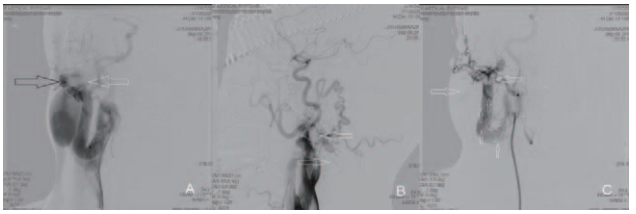

FIGURE 5. Angiograms in embolization. (A) Anteroposterior angiogram revealed the first no. 2 detachable balloon (white arrow) in the orificium fistulae (black arrow). (B) Lateral angiogram revealed the second no. 2 detachable balloon (white "←") in the orificium fistulae and the metallic ball of the first broken balloon (white "→") in the vein lake. (C) Anteroposterior angiogram revealed the third balloon (white "←") in the orificium fistulae and Interlock-35 coils (white "↑") in the U-shaped artery loop, and the diameter of U-shaped artery loop was markedly wider than the inflated no. 2 detachable balloon.

maxillofacial region gradually relieved 2 weeks later, during this period lente liberantes analgesics were given once a day and oppression to right CCA was given 4 times in the first postoperative week. Computed tomography angiography (CTA) revealed little contrast media in ascending branch of U-shaped loop and orificium fistulae was out of visualization (Fig. 7). He experienced no postoperative neurologic deterioration and hyperperfusion syndrome and was discharged in soundly independent status 4 weeks after the second therapy. Four months later, control angiograms confirmed that the AVF was completely occluded, and no other

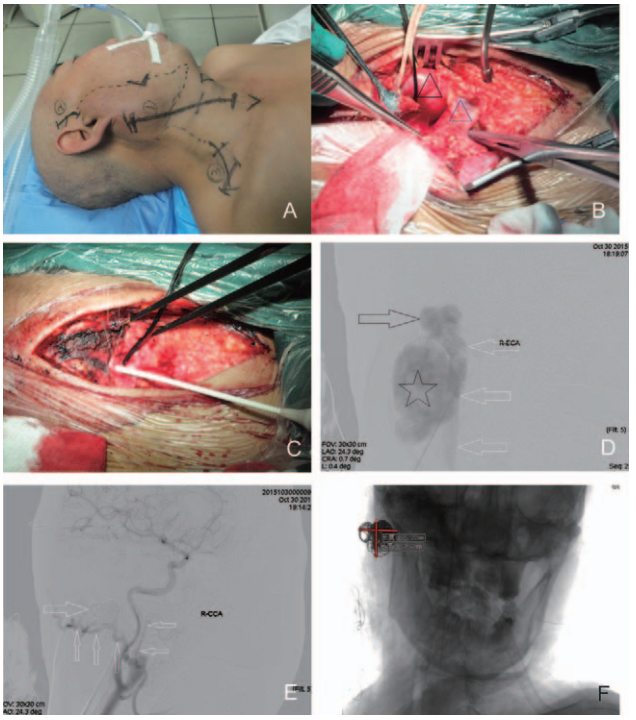

FIGURE 6. Photograms and angiograms in the second therapeutic procedure. (A) Photogram revealed where these 3 incisions were made and the posture. (B) Photogram revealed the proximal part of common facial vein (blue "△") and the lowest segment of U-shaped loop (black "△"), these 2 vessels were all revealed for ligation. (C) Photogram revealed a 6F sheath, used for embolism and contrast examination, was implanted into the ascending portion of U-shaped arterial loop at centrifugal side of ligating silk suture (↓). (D) Angiogram revealed the orificium fistulae (→), ascending portion of U-shaped arterial loop (←), and venous lake (☆). (E) Angiogram clearly revealed internal carotid artery, occipital artery (↑), ascending pharyngeal artery (←), and the mass of coils (white →) in the orificium fistulae, where no contrast agent entering and that all of the feeding arteries and draining veins were soundly disappeared.

FIGURE 7. (A) Computed tomography angiography revealed little contrast media in ascending branch of U-shaped loop while orificium fistulae is out of visualization, and there was only one effective ligature in the loop. (B) The mass of detachable coils was over the neck of condyle.

FIGURE 8. (A, B) Angiograms revealed that the arteriovenous fistula was completely occluded, ICA showed clear visualization, and no other lesion was found.

lesion was found (Fig. 8). The patient was contacted 12 months later, there was a hard mass, about thumb tip-sized, existing prior to right tragus, and his other symptoms all had resolved.

DISCUSSION

Traumatic AVF in preauricular region is fairly rare.[5,10] Most of these fistulae occur as a result of an incidental injury[11] or iatrogenic injury[1,3–9] (Table 1). Spontaneous occurrence in maxillofacial region has also been reported.[12,13] Difference trauma, a blunt or sharp trauma, to local jacent blood vessels may immediately induce AVF or indirectly facilitate the formation of AVF.[4,5,8,11,14]

The suppositional mechanism is anatomic difference that the soft subcutaneous tissues in preauricular region is more mobile and thicker than that in scalp and there is no smooth outer table of the skull in preauricular region. So the preauricular region is not particularly susceptible to blunt injury as scalp. To this patient we supposed the development of the AVF in preauricular region was from the direct puncturing injury to the main trunk of STA and adjacent veins.

With regard to the mechanism of formation of post-traumatic AVF in preauricular region, at least 2 mechanisms have been suggested.[5,15–17] One is the laceration theory that simultaneous lacerations of the artery and the accompanying vein result in a single fistula.[1,4,5,8,10,11,14,18] The other is the disruption theory of the vasa vasorum of the arterial wall, which proposes that endothelial cells proliferating from the vasa vasorum into the hematoma around the disrupted vasa vasorum form endothelial buds and numerous small vessels[1,19] resulting in numerous vascular channels created to adjacent veins. Concerning the mechanism of the lesion in our patient, according the history of pricking injury from "flying cutter" and the outcome of auxiliary examination, the STA main trunk and the neighboring venous systems maybe simultaneous directly or indirectly injured, and the AVF generated and gradually grew along with time. As the fistula increases in size, additional peripheral vessels contribute to the high-flow, high-pressure shunt.[5,11] The natural history of traumatic AVF is not completely understood. Most patients with traumatic AVF in scalp, head and neck were found in the period of about a few days to months postoperatively or after trauma.[5,15,18] This patient was symptomless in the first 20 years after prick and the symptom tardily developed in the subsequent 20 years, it may be explained that the factors such as dysvascular status, hemodynamic specialty of regional blood vessels, and anatomic features adjacent anatomic structure influenced the onset and development of the fistula.[6]

The indication of therapy includes improvement appearance, elimination of symptom such as tremor and tinnitus, and reduction the risk of health lesions such as hemorrhage and steals flow. Two categories fundamental therapeutic methods, chirurgic surgery and transvascular interventional therapy have constantly been used to deal with these lesions, the former including excision of all the fistula and ligation of relative vessels has been the traditional therapeutic modality of dealing with such lesions in vast majority in the past and now is still constantly being adopted,[20,21] the latter including transarterial and transvenous embolization has been a gradually prevalent essential therapeutic modality for its rapid, safe, and highly effective virtue.[1,13,22–24] The therapy of injection of sclerosant[25] in focal zone or radiation therapy[12] were also adopted. Simple therapeutic method maybe effective for simple patient,[7] but for large and complicated AVF may need combined treatment.[26] The lesion in the present patient was a large, complicated, high-flow AVF whose orificium fistulae around the right neck of condyle, that impeded surgical access to exposure and excision.[27] Embolization has been proposed as a safer alternative to surgical ligation or resection in such situations.[7,27,28] We first applied transarterial embolization with detachable balloons and ciliary spring coil but

TABLE 1. List of Reports

Year	Authors	No of Patient	Location	Cause	Complicated AVF*
1991	Preisler et al[4]	1	Superficial temporal artery	Iatrogenic	No
1991	Lanigan et al[5]	1	Internal maxillary artery	Iatrogenic	No
1997	Scholl and Rutledge[6]	1	Uperfacial temporal artery	Iatrogenic	No
1999	Calwell et al[7]	1	Uperfacial temporal artery	Iatrogenic	No
2004	Martin-Granizo et al[8]	1	Uperfacial temporal artery	Iatrogenic	No
2013	Janssen et al[1]	1	Superficial temporal artery	Iatrogenic	No
2014	Sacho et al[9]	1	Superficial temporal artery	Iatrogenic	No

AVF, arteriovenous fistula.
*The AVF with more than 1 supplying arteries and draining veins.

failed after effort of 4 hours. At first, the coils could be partly coiled in the orificium fistulae but we could not release it for its instability under the vigorous impact force of blood stream even we abated it through compression. And we could not release the balloon because the orificium fistulae was too large that there was no enough frictional resistance between it and the balloon for detachment. We also could not use PTA Balloon Dilatation Catheter (Bard Peripheral Vascular, Murray Hill, NJ), which has a straight balloon of 4 cm—length can be inflated to the diameter of 12 or 16 mm according its type, to obstruct blood stream for there is a short 180° N-shaped loop at the head of ECA. It is critical to select the suitable embolic material that match the size of the shunt for success in embolotherapy,[5,22] but we found it was difficult for such a big AVF at that time. The patient was at last swimmingly cured through combined therapy of chirurgic ligation of feeding arteries, draining veins and embolization of orificium fistulae via transarterial path. In the therapeutic procedure, the powerful blood stream in the orificium fistulae was successfully blocked by ligating the feeding arteries, which made it feasible to steadily embolize the orificium fistulae with unmixed detachable coils. In fact the orificium fistulae was not tightly embolized with unmixed detachable ciliary coils, we concluded that the constriction of soft tissues after ligation of feeding arteries may have further reduced the relict hematologic supply to the orificium fistulae, and once the fistula has been obliterated by thrombosis the occlusion would be stable.[22] The main purpose of ligating the common facial vein and the external jugular vein at their centripetal distal ends and postoperative use of low molecular heparin was to avoid the formation of antidromic thrombus in the stumps of these grossus veins. The treatment protocols for such a formidable and high-risk patient, in our experience, should be made according the character of the extensional lesion[8,13] after extensive discussion among related professional specialists,[29] and the combined therapeutic method maybe the practical alternative.

CONCLUSION

Large and complicated traumatic AVF in preauricular region is rare, often due from a injury in maxillofacial region, combined therapy is needed. How such lesions should be managed on an individualized basis and extensive discussion is especially helpful in making suitable treatment protocols for such a formidable and high-risk patient.

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