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Case Report

The perplexing postsurgical complication of carotid-jugular fistula: A bitter experience

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ABSTRACT

Background: Vascular injuries occur in approximately 25% of all penetrating neck traumas, with carotid artery injuries being particularly lethal. Penetrating neck injuries are potentially fatal. Vascular injuries occur in approximately 25% of cases, which can lead to the formation of arteriovenous fistulas.

Case Description: The authors present a case of delayed open surgery to repair a carotid-jugular fistula that resulted in an unprecedented complication, as well as a brief review of the condition's diagnosis and treatment options.

Conclusion: This case report suggests us that, penetrating neck injuries should be thoroughly evaluated for arteriovenous fistulae. To avoid complications, common carotid-jugular fistulas must be treated as soon as possible. Postoperative complications can be effectively managed with prompt action.

Keywords: Arteriovenous fistulas, Carotid-jugular fistula, Neck injury, Neurosurgery, Neurotherapeutics

INTRODUCTION

Approximately 25% of all penetrating neck traumas result in vascular injuries, and carotid artery injuries are particularly lethal.[10] Any penetrating trauma to the neck with vasculature injury may result in the formation of pseudoaneurysms (PSA) and associated venous injury may result in an arteriovenous fistulas (AVF). Despite its rarity, carotid-jugular fistula (CJF) accounts for nearly 4% of all traumatic AVFs.[6] Hunter first observed and described the pathophysiology of AVFs in 1757, and published a report on it in 1764.[1,6] Long after that, in 1951, Warren *et al.* described a posttraumatic CJF.[14] AVFs in the neck region can be congenital or acquired, with the majority of AVFs being acquired and having an accidental or iatrogenic traumatic origin, necessitating early intervention.[5] Congenital fistula between the carotids and the internal jugular vein is uncommon and can be clinically silent for a long time.[2]

CASE REPORT

A 35-year-old male presented to us with a history of a stab injury to the right side of his neck with profuse bleeding 2 months before admission. Following the injury, he lost consciousness for 10 min and developed visual disturbance in the form of the left homonymous inferior quadrantanopia. He was neurologically intact and had GCS 15 at the time of presentation. An ill-defined cystic and compressible swelling just below the angle of the mandible was discovered on examination of the neck, which had a well-healed scar mark over it. The swelling had a palpable thrill and bruit that coincided with the apex beat. However, no abnormalities in cardiac status were discovered.

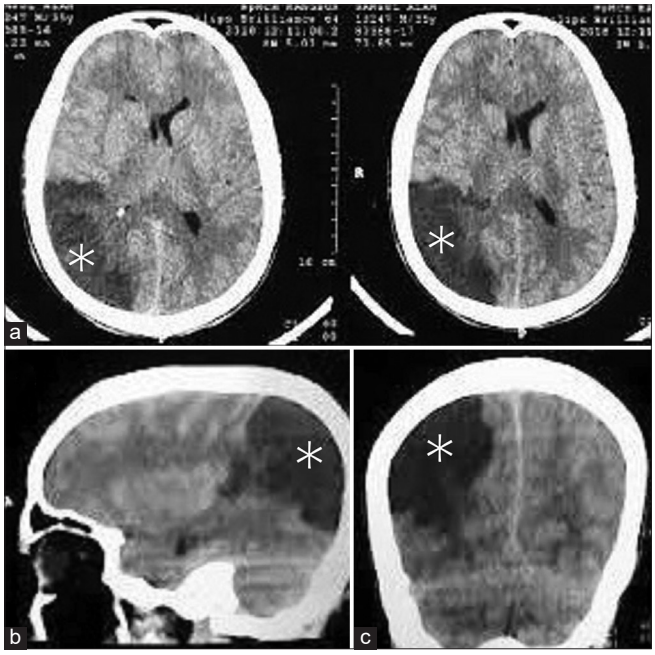

Figure 1: Pre-operative CT scan of the patient showing right parieto occipital infarction.

Although the patient had no significant neurological morbidity, a CT scan of the brain revealed a large right parieto-occipital infarction [Figure 1], while a computed tomography angiography (CTA) revealed a PSA at the bifurcation of the right CCA that communicated with the IJV. On the right side, the external carotid artery (ECA) and its branches were not visible [Figure 2]. Digital subtraction angiography (DSA) revealed a large fistula between the CCA bifurcation site and the IJV, with blood being pulled from the right ICA to the IJV through the fistula tract [Figure 3a]. The anterior communicating (A-Com) and posterior communicating (P-Com) arteries provided adequate cross circulation [Figures 3b and 3c]. Because the fistula was so strong, it drew blood from the opposite posterior circulation [Figure 3d].

The surgery was planned to remove the PSA and repair the right ICA and IJV to reconnect the fistula. The PSA, measuring about 5 cm × 3 cm, was discovered just a short distance from the start of the ICA, and the fistula was discovered to be of such a large size that the surgical plan had to be revised intraoperatively. Bypass was also not an option, and because the left ICA fed the right cerebral hemisphere very well through the right MCA, the right ICA was ligated just above and below the connection with the PSA to trap it. Fistula ligation was also attempted, though due to its large caliber, only a ligature was attempted, and full obliteration was not possible. The PSA was discovered to originate at the junction of the CCA and ICA and was inseparable from the IJV, which was severely dilated.

The patient was neurologically intact until the 2nd postoperative day (POD). On the 3rd POD, the patient suddenly deteriorated and became left hemiplegic with MRC Grade 0 in both the upper and lower limbs. GCS also dropped

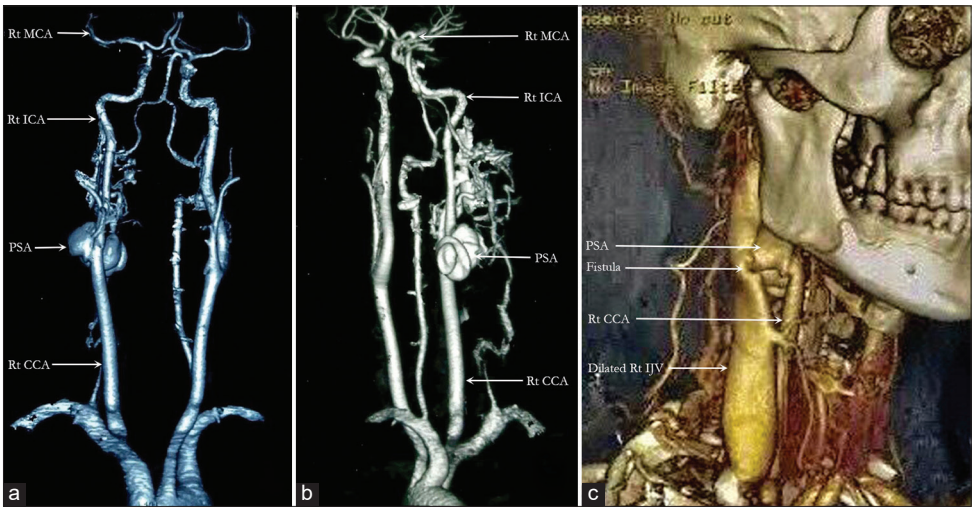

Figure 2: Pre-operative computed tomography angoigraphy showing pesudo aneurysm at the bifurcation of right CCA.

to 11, and the right pupil became dilated and nonresponsive to light. In addition to the previous right parieto-occipital infarct with significant midline shift, a CT scan of the brain revealed a large right-sided MCA territory infarct [Figure 4]. CTA revealed a reduction in the size of the PSA while the fistula remained patent and the right MCA was not visible beyond its origin [Figure 5]. A right fronto-temporo-parieto-occipital decompressive craniectomy (DC) was performed right away. The brain was found to be swollen, tight, and nonpulsatile postoperatively.

On the 2nd post discharge POD, GCS increased to 15, and the power of the left lower limb improved to MRC Grade 2 while the power of the left upper limb remained at MRC Grade 0. The right pupil was 6 mm in diameter and did not respond to light, whereas the left pupil was 4 mm in diameter and did respond to light. After 4 weeks of DC, cranioplasty was performed using autologous bone. After another 4 weeks of DC, the patient was discharged with MRC grades of 3 and 2, respectively, in the lower and upper limbs.

At 6 months, the patient's left lower and upper limb power had improved to MRC Grades 4 and 3, respectively, and he could walk with assistance. However, the homonymous quadrantanopia remained unchanged.

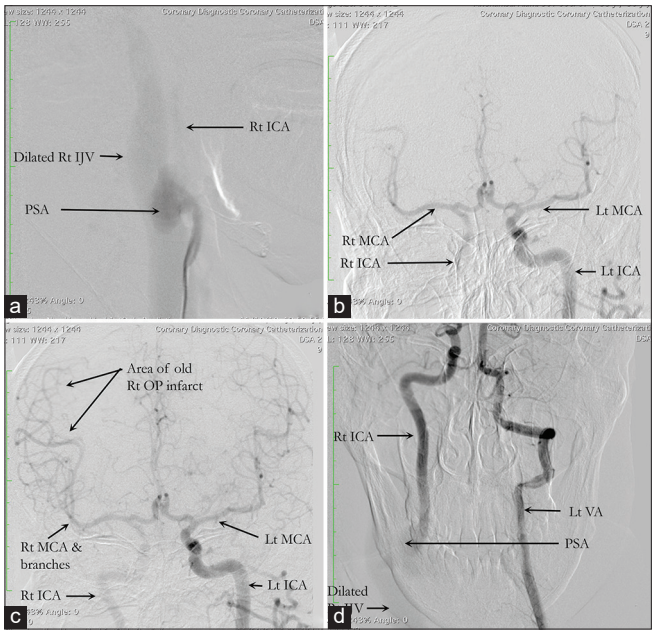

Figure 3: Pre-operative DSA showing large fistula between CCA bifurcation site and IJV(A)., anterior and posterior communicating artery providing adeuate cross circulation (3B, 4C). Fistula also taking blood from opposite posterior circulation (3D).

DISCUSSION

AVF is an abnormal communication between an artery and a vein that occur as a result of penetrating trauma to both. An AVF is primarily a shunt that allows blood to flow from the arterial system with the higher pressure to the venous system with the lower pressure while bypassing high resistance arterioles.[1] CJF, an anomalous communication between the carotid artery and the jugular vein, is a relatively uncommon AVF. These abnormalities may be congenital or acquired. Congenital fistulas, which are much less common than acquired fistulas, are usually associated with collagen vascular disease, whereas acquired fistulas are mostly the result of penetrating trauma, both accidental and iatrogenic, or even blunt trauma may rarely give rise to AVFs.[4,13]

Penetrating injuries cause the majority of traumatic AVFs in the neck. Stab wounds, followed by gunshot wounds, are the most common modes of accidental injury that result in CJFs iatrogenic injury during the central venous catheter insertion of the IJV has recently been found to cause more CJFs than accidental injuries.[1,9,11] Congenital AVFs are rare and result from persistent communications between developing arteries and veins during embryonic life, which may be triggered by menarche, pregnancy, or collagen vascular disease.[2,4] The CJF in our patient was traumatic, resulting from the most common mode of injury, stabbing.

Traumatic AVFs are frequently missed during the acute phase of injury, resulting in treatment being initiated weeks

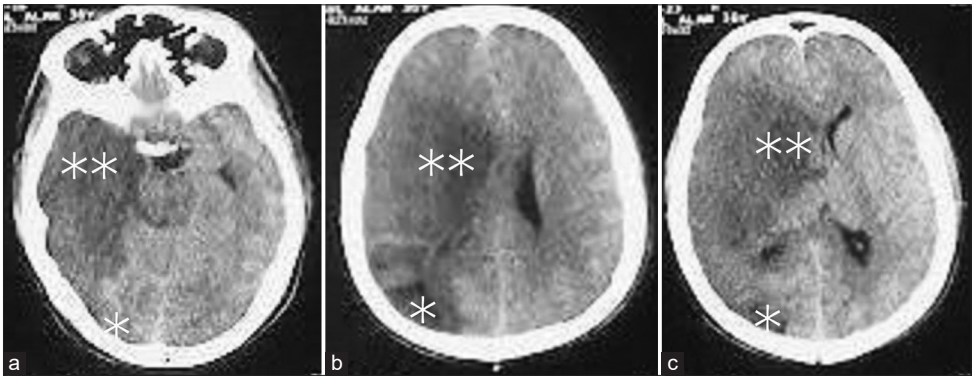

Figure 4: Post operative CT scan showing large right sided MCA territory infarct.

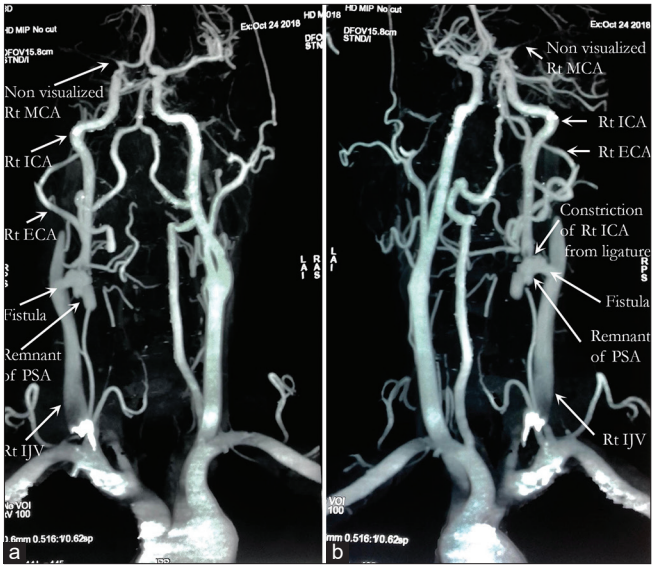

Figure 5: Post operative CTA revealed reduction in size of pseudo aneursysm.

or even months after the initial trauma.[8] These vascular fistulae commonly present clinically as pulsatile swelling with thrill and bruit, edema, and dilation of superficial veins in the neck. The Nicoladoni-Branham sign, which is characterized by bradycardia and elevated mean blood pressure in response to physical compression of the fistula, can be seen in some patients. If not treated, they can cause complications such as rupture and bleeding, infection, tinnitus, arrhythmias, congestive cardiac failure, steal phenomenon, or thromboembolism, which can impair cerebral circulation and cause fainting episodes or ischemic stroke.[2,5-7,9,11,12] Our patient presented 2 months after the incident. It was most likely a late presentation because there were no neurological or other deficits other than left homonymous inferior quadrantanopia. This visual deficit was most likely caused by a right parietal infarct that he had long before the injury. It is possible that the patient had this quadrantanopia for a long time and was adopted with it, and thus did not notice or complain about it until it was discovered only after a thorough examination. He only came to us when the swelling in his neck became noticeable and bothersome. Despite the fact that he presented at a late stage, he did not develop any cardiac manifestations.

To make a diagnosis of traumatic AVF and PSA, a careful history and physical examination are required. Nonetheless, additional noninvasive and invasive studies are required for definitive diagnosis and intervention planning, whether endovascular, conventional, or hybrid surgery are used.[6,12]

The investigations of choice are CTA, magnetic resonance angiography (MRA), Doppler ultrasonography, and DSA. All of these can identify the feeders and drainers, as well as the precise location and size of the fistula. Because of their noninvasiveness, CTA and MRA have largely replaced DSA in recent years. However, DSA has the added benefit of being able to perform interventional therapy at the same time.[1,5,6] We started with CTA, which revealed all of the vessels up to the MCAs on both sides except the right ECA, fistula, and PSA [Figure 2]. We performed a DSA to see the flow pattern and were pleasantly surprised to find the high-flow fistula pulling blood from the left anterior circulation through the A-Com and even from the posterior circulation through the P-Com while maintaining adequate cross-circulation to fill the right MCA very well [Figure 3].

The neck is particularly vulnerable to external trauma, and its unparalleled intricate anatomy of close proximity of important neurovascular and aerodigestive structures makes the surgical approach to any pathology in the cervical region particularly difficult.[10]

Surgical intervention, whether endovascular, conventional, or hybrid, is the treatment of choice. The anatomy, size, and shape of the fistula and/or PSA, extent of the initial arterial injury, distal arterial flow, and facilities available should all be considered when deciding on a procedure.[6]

Repairing AVFs should be attempted as soon as possible if there is hemodynamic instability or evidence of vascular injury. Late surgery becomes difficult due to fibrosis and collateral development because the anatomy becomes distorted, and dissection becomes risky. The risk of hemorrhage may also increase with late surgery. Both AVFs and PSAs can be effectively treated with either traditional surgery or endovascular techniques.[4,6]

The primary goal of CJF management is to close the fistula and reconstruct the involved vessels. In general, open repair of a CJF by conventional surgical intervention includes end-to-end anastomoses, repair with patches, interposition grafts, or damage control procedures in some unavoidable situations. When reconstruction is impractical and the situation calls for it, ligation of the AVF's contributing vessels should be considered. Endovascular techniques outperform surgical dissection, especially in cases of anatomic distortion, such as large AVFs associated with PSA, where they can be manipulated more easily than open surgery.[1,4,6,10,11]

Endovascular techniques for CJFs that are currently available include embolization with a balloon, coil, or glue, as well as the placement of covered stents or self-expandable graft stents. Advances in the development of biodegradable stents allow for the avoidance of stent-related complications. Endovascular therapy is a better choice of management because it has fewer complications than open surgery. Endovascular therapy also has less invasiveness, a lower risk of hemorrhage, and a shorter hospital stay. Furthermore, this allows for the selective occlusion of the fistula, which

is strongly advised to avoid further complications.[2,4-7,11] Nonetheless, due to the scarcity of incidences, case series, or long-term follow-ups, particularly regarding prognosis and complications, there are insufficient data to judge the superiority of open versus endovascular surgery. Despite the fact that the majority of the literature has shown that both procedures have a high success rate.[3,4] Given the available resources, we chose and planned for open surgery to excise the PSA and repair of the right ICA and IJV to close the fistula. Nonetheless, operative findings and circumstances led us to deviate from the preoperative plan and trap the right ICA by ligating it just above and below the PSA. The fistula was also attempted to be ligated, but due to its large size and the adhesion of its thin wall to the surrounding structures, effective ligation was not possible. Following surgery, the PSA was significantly reduced in size, and the right CCA, which could not be seen in preoperative CTA and DSA, became visible, though the caliber was not as large as the left one. The CCA reappeared, most likely as a result of a decrease in the flow into the fistula [Figure 5].

Because most CJFs can be effectively managed through open surgery, endovascular procedures, or a combination of the two, postsurgical complications have received little attention in the literature. The possibility of disaster from intraoperative blood loss, as well as injury to vital neurovascular and other surrounding structures, raises the risk of death from open surgery. Endovascular procedures, on the other hand, may increase the risk of thromboembolism, late intrastent stenosis, or even the formation of PSA at the site of arterial puncture.[2,5,6] Endovascular procedures for traumatic AVFs and PSAs, on the other hand, may increase the risk of thromboembolism, late intrastent stenosis, or even the formation of PSA at the site of arterial puncture.[6] Nonetheless, the type of complication that we encountered had not previously been described in the literature. The hemiparesis with the lower GCS in our patient was most likely caused by decreased flow to the right MCA, which resulted in an infarct in the MCA territory due to less circulatory volume from the opposite ICA or the posterior circulation, as the pulling effect of the fistula became much less postoperatively, as seen on postoperative CT and CTA [Figures 4 and 5]. Furthermore, the patient's ambulation may have altered his hemodynamics, adding insult to injury. Prompt DC most likely saved the penumbra, allowing the patient to gradually regain motor power.

CONCLUSION

Penetrating neck injuries should be thoroughly evaluated for arteriovenous fistulae. To avoid complications, the common CJFs must be treated as soon as possible. Postoperative complications can be effectively managed with prompt action.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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Commentary

The authors present an unusual case of traumatic carotid-jugular fistula of high enough flow to cause a steal phenomenon. They should be commended for presenting their case as a cautionary example of a severe complication related to carotid sacrifice. It is always easy to be critical in retrospect, but previous work has shown that despite what would appear to be good collateral circulation on imaging studies, some patients will fail to tolerate carotid sacrifice, and the consequences can be significant. Safer options would have included an endovascular approach with ICA preservation, some form of cerebral revascularization to protect the hemisphere, trapping of the fistula with direct repair, or sacrifice of the internal jugular vein.

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