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CASE REPORT

CLINICAL CASE

Traumatic Common Carotid-Internal Jugular Arteriovenous Fistula Manifesting as Life-Threatening Epistaxis



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ABSTRACT

Post-traumatic common carotid artery-internal jugular vein fistula is extremely rare (4% to 7% of all traumatic arteriovenous fistulas). Clinical manifestations depend on the size, duration, and proximity to the heart. This report describes a case where the condition remained undiagnosed for years, and the patient was recently treated with transsternal ligation of the left common carotid artery. (Level of Difficulty: Advanced.) (J Am Coll Cardiol Case Rep 2019;1:576-8) © 2019 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

raumatic common carotid artery-internal jugular vein fistula (CJF) is a rare entity with a published incidence of only 4% to 7% of all traumatic arteriovenous fistulas. It is caused by penetrating injury or by internal jugular venous

LEARNING OBJECTIVES

- Post-traumatic CJF is an extremely rare clinical condition. Timely treatment is essential to avoid development of complications.
- This may be missed during the acute phase of injury, hence one should have highest degree of suspicion.
- All such injuries should be evaluated by computed tomography angiography to establish or rule out the diagnosis.
- The treatment should be individualized on the basis of the anatomy of the lesion and the safety of the available treatment.
- With a careful work-up and individualized intervention, this condition has a good clinical outcome.

cannulation, and it may remain untreated for months or years after the initial injury (1). If it is left untreated, the patient is likely to develop high-output cardiac failure, embolization, and/or atrial fibrillation. We report a case that remained undiagnosed for 20 years and was successfully treated by transsternal ligation of the left common carotid artery, a procedure rarely reported in publications. Direct surgical repair of the fistula was not a safe option, nor was the endovascular approach feasible in this case.

HISTORY OF PRESENTATION

A 60-year-old man was referred to our hospital (Holy Family Hospital, New Delhi, India) after 4 episodes of nasal bleeding and 1 episode of transient loss of consciousness. He had also experienced a progressive increase in breathlessness on moderate exertion and decreased effort tolerance in the preceding 3 years. Clinical examination was remarkable for a pulsatile soft tissue swelling along with prominent veins on the left side of his neck, an old scar, a palpable thrill, and a harsh murmur over the swelling. Branham's

Informed consent was obtained for this case.

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sign was positive. The examination of other systems was unremarkable.

PAST HISTORY

The patient had experienced an arrow injury on the left side of his neck 20 years previously and had undergone a surgical wound exploration along with multiple blood transfusions at another institution.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included aneurysm, hemangioma, and arteriovenous malformation.

INVESTIGATIONS

Color duplex ultrasonography revealed a communication between the left common carotid artery and the internal jugular vein. In addition, magnetic resonance carotid angiography revealed a fistulous communication (3.71 mm in diameter) between the left common carotid artery and the adjacent internal jugular vein with total occlusion of the left common carotid artery beyond the fistula. Further, there was a small aneurysm at the site of the fistula. The internal jugular vein was hugely dilated, and the left vertebral artery was very prominent (Figures 1 and 2).

MANAGEMENT

The endovascular approach was not a therapeutic option for technical reasons (see Discussion). Because the left common carotid artery was occluded beyond the fistula and the fistula was situated in difficult zone I, a safer approach was chosen, and the left common carotid artery was ligated at its origin through sternotomy. The thrill disappeared, and the patient's post-operative recovery was uneventful. At 1-year follow up, he was asymptomatic, and there was remarkable reduction in the swelling and prominence of the neck veins.

DISCUSSION

Traumatic common carotid-jugular fistula is extremely rare, and its rarity can be gauged from its appearance only as case reports in medical publications (2). A total of 24 cases were reported by 22 authors from 2000 to 2012. Of these, only 9 of these fistulas were caused by a gunshot or stab injury; the remaining resulted from internal jugular vein catheter insertion (2). A few of the cases were diagnosed quite late, as in our patient. Kakkar et al. (3) reported a case of CJF lasting 28 years that was surgically treated after the patient had repeated hospitalizations for control of congestive heart failure. However, the longest presentation of CJF was 58 years after a gunshot injury; the CJF was then treated by surgical repair (2). Besides the duration, the severity of symptoms is largely determined by the fistula's characteristics,

mainly its size and proximity to the heart. A largediameter (>8 mm) fistula leads to early high-cardiac output failure, but a small-diameter (<5 mm) fistula may manifest many years after injury, as in our case. The episode of nasal bleeding in our patient case was secondary to rupture of an engorged nasal vein, and transient loss of consciousness may have been caused by a transient ischemic attack from the right carotid artery or the posterior circulation.

The endovascular option was not possible because of unfavorable anatomic features, mainly the absence of a landing zone and the presence of an aneurysm. Moreover, direct surgical repair was not considered safe because of the location of the fistula in the difficult zone I, the presence of surrounding scar tissue, and hugely dilated veins from the resultant venous hypertension. The lateral neck is divided into 3 zones. Zone I extends from the clavicle to the cricoid cartilages, zone II from the cricoid cartilages to



Short T₁ inversion recovery long T₁ image showing a void of flow-related signal in both the left common carotid artery (LT. CCA) and the internal jugular vein (IJV). A = anterior; F = foot; H= head; L = left; R = right.

ABBREVIATIONS AND ACRONYMS

CJF = common carotid arteryinternal jugular vein fistula FIGURE 2 Magnetic Resonance Angiogram



The image shows the common carotid artery-internal jugular vein fistula, the hugely dilated internal jugular vein (IJV), the occluded left common carotid artery (LT. CCA) beyond the fistula, and a very prominent left vertebral artery (LT. VERTEBRAL ART.). Abbreviations as in **Figure 1**.

the angle of the mandible, and zone III from the angle of the mandible to the base of the skull. It is difficult to achieve adequate exposure for fistula repair in zones I and III, whereas adequate exposure it is relatively easier to achieve in zone II. Considering all these factors, ligation of the left common carotid

because there was no requirement for maintaining vascular continuity. The result was gratifying. Ligation of the left common carotid artery has rarely been used and reported (1). Sometimes repair of this type of fistula may be complex enough to require cardiopulmonary bypass (4). In 2004, a paradigm shift in the treatment of CJF occurred when Droll and Lossing (5) performed the closure using a covered stent in which the vessel distal to the fistula was patent. Whether the approach to treatment of a CJF should be endovascular or surgical is debatable, and this determination is still made on a case-by-case basis (2). However, the primary indications for surgery are a large fistula, an associated aneurysm, the presence of the fistula in zone II, and/or the involvement of other vessels.

artery at its origin was considered to be very safe

FOLLOW-UP

At 1-year follow-up, the patient was asymptomatic, and there was no visible swelling, thrill, or murmur at the site of the previous malformation.

CONCLUSIONS

Post-traumatic CJF is an extremely rare clinical condition that may be missed during the acute phase of injury. All penetrating neck injuries should be carefully evaluated by computed tomography angiography to confirm the presence or absence of CJF. The therapeutic strategy of a long-standing fistula should be individualized depending on the anatomy and situation of the lesion, as well as the safety of the available treatment choices.

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REFERENCES

1. Ezemba N, Ekpe EE, Ezike HA, Anyanwu CH. Traumatic common carotid-jugular fistula: report of 2 cases. Tex Heart Inst J 2006;33: 81-3.

2. Caldarelli C, Biricotti M, Gabriele M, et al. Acquired carotid-jugular fistula: its changing history and management. ISRN Vasc Med 2013;2013: ID320241. **3.** Kakkar S, Angelini P, Leachman R, Cooley DA. Successful closure of post-traumatic carotid-jugular arteriovenous fistula complicated by congestive heart failure and cerebrovascular insufficiency. Cardiovasc Dis 1979;6:457-62.

4. Talwar S, Bhan A, Chaudhary SK, et al. Carotid artery to jugular vein fistula. Asian Cardiovasc Thorac Ann 2000;8:366–8.

5. Droll KP, Lossing AG. Carotid-jugular arteriovenous fistula: case report of an iatrogenic complication following internal jugular vein catheterization. J Clin Anesth 2004;16:127-9.

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