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Case Report

Trigeminal neuralgia due to intracranial venous reflux following central venous disease in a patient on hemodialysis: A case report

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ABSTRACT

Background: A wide variety of conditions can cause trigeminal neuralgia (TN).

Case Description: We describe a rare case of a 77-year-old female patient on hemodialysis presenting with severe TN on the right side of the face for several weeks. She underwent multiple revisions using catheter for brachiocephalic venous stenosis over 6 years after a therapeutic arteriovenous fistula (AVF) was created in the left forearm. Her facial pain was consistent with Type 1 TN and remained intractable even after carbamazepine treatment. The initial magnetic resonance imaging did not demonstrate arterial compression on the right trigeminal nerve; instead, the vein adjacent to the right trigeminal nerve showed a hyperintense signal. In addition, the contralateral cortical veins and transverse sigmoid sinus were dilated. Angiography from the left brachial artery revealed intracranial venous reflux (IVR) through the left jugular vein due to an occluded brachiocephalic vein. Her pain was relieved immediately after her left upper arm was compressed with a sphygmomanometer to decrease the shunt. Surgical elimination of the AVF on the left forearm resulted in complete resolution of TN. Postoperative radiological examination revealed the resolution of IVR, and her TN has not recurred by her 6-month follow-up.

Conclusion: The radiological diagnosis of IVR might be complicated because the true causative lesion for focal neurological symptoms might be remotely located. IVR following central venous disease should be a differential when patients on hemodialysis present neurological symptoms.

Keywords: Central venous disease, Hemodialysis, Intracranial venous reflux, Trigeminal neuralgia

INTRODUCTION

A wide variety of conditions can cause trigeminal neuralgia (TN). Arterial compression accounts for 80% of all vascular compression observed in patients with Type 1 TN, which is characterized by the following: electric shock pain, trigger point, and a certain degree of carbamazepine effect.[12] The therapeutic effect of surgery is well established for cases caused by arterial compression; microvascular decompression cures the disease in 85% of cases.[14] In contrast, no surgical strategy has been established for cases without obvious arterial compression.[2] We present a case of TN without arterial compression in a patient on hemodialysis. Based on imaging studies, intracranial venous reflux (IVR) due to the central venous disease (CVD) was suspected

as a causative mechanism. Since CVD is relatively common with a reported risk of approximately 30% in patients on dialysis,[1,9-11] we believe that IVR due to CVD should be considered when patients on dialysis present with TN with no evident arterial compression.

CASE DESCRIPTION

Patient history and examinations

A 77-year-old woman presented with a 3-week history of severe right-sided facial pain. The patient's pain was characterized as electric shock-like pain, lasting 1–2 s, and triggered by touching the face. She had a 6-year history of end-stage renal disease managed by hemodialysis using an arteriovenous fistula (AVF) in her left forearm. Recently, the AVF in the left forearm required multiple percutaneous transluminal angioplasty (PTA) procedures because of intractable stenosis within the AVF. Stenosis at the brachiocephalic vein was also detected and was treated with PTA. One month before visiting our hospital, another AVF was newly placed in her right upper arm. She presented with no other neurological complaints apart from facial pain. Moreover, she had initially received carbamazepine with no relief of pain.

Although the characteristics of her facial pain were consistent with typical Type 1 TN, magnetic resonance imaging (MRI) indicated no apparent arterial abnormalities near the right trigeminal nerve [Figure 1a]. However, time-of-flight (TOF) magnetic resonance angiography (MRA) showed an abnormal high-intensity signal in the vein adjacent to the right trigeminal nerve [Figure 1b] and the cortical veins and transverse sigmoid sinus on the left side were dilated [Figure 1c]. These findings suggested an IVR attributed to the AVF in the left forearm. Angiography from the left brachial artery revealed occlusion of the brachiocephalic vein and retrograde flow to the cranial venous system through the left jugular vein and lateral sinus, presenting with dilated cortical and diploic veins [Figure 2a]. The flow ultimately drained through the right jugular vein [Figure 2b]. The remaining AVF in the left forearm was subsequently removed under local anesthesia.

The postoperative course was uneventful, and her TN resolved immediately. Postoperative MRI showed the resolution of both the IVR [Figure 2c] and the abnormal signal adjacent to the right trigeminal nerve [Figure 2d]. Postoperative angiography revealed occlusion of the left jugular vein and anterograde flow at the left transverse sigmoid sinus, which subsequently drained into the right venous system through collateral veins. The patient had no facial pain at the 6-month follow-up. Written agreement for publication was obtained from the patient.

DISCUSSION

The present case indicates that it is essential to search not only for the subtle findings around the trigeminal nerve but also the entire head-and-neck region for rare causes of TN, especially in the absence of arterial compression. In this case, IVR due to CVD after the therapeutic AVF in the left forearm appears to have caused dilatation and increased vessel pressure in the transverse pontine vein adjacent to the trigeminal nerve. The fact that her facial pain immediately resolved after a simple bed-side maneuver to compress the left forearm using a sphygmomanometer and that an abnormal signal on TOF-MRA disappeared after surgical closure of the shunt supported our speculation.

CVD is characterized by either stenosis or occlusion of veins and is a well-known chronic pathological condition in patients on dialysis.[1,9-11] The precise mechanism of CVD in patients on dialysis remains controversial, but it is speculated to result from mechanical endothelial damage and subsequent inflammation at the initial insertion of the

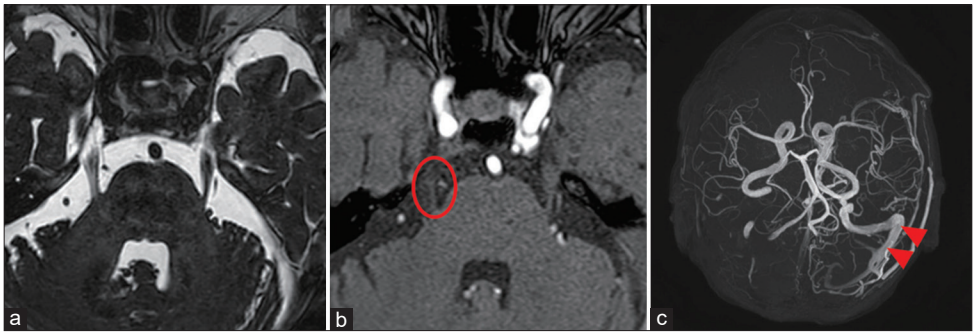

Figure 1: (a-c) Preoperative magnetic resonance (MR) imaging. (a): An MR constructive interference in steady state image showed no arterial compression on the right trigeminal nerve. (b) A time-of-flight MR angiography demonstrated a slightly abnormal high-intensity signal from a vein adjacent to the right trigeminal nerve (red circle). (c) A maximum-intensity projection MR angiography revealed a retrograde venous flow in the left transverse and sigmoid sinus (red arrowhead).

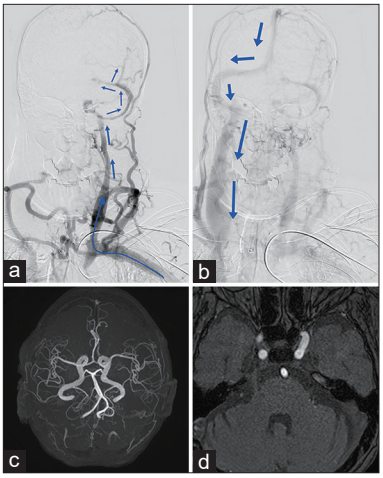

Figure 2: (a) A preoperative angiography from the left brachial artery revealed an occlusion of the brachiocephalic vein and retrograde flow to the cranial venous system through the left jugular vein and lateral sinus (blue arrows), causing dilated cortical and diplopic veins. (b) The late phase of the angiography showed that the flow ultimately drained through the right jugular vein (blue arrows). (c) A postoperative maximumintensity projection magnetic resonance (MR) angiography demonstrated the disappearance of the retrograde venous flow in the left transverse-sigmoid sinus. (d) The abnormal high-intensity signal adjacent to the right trigeminal nerve on the time-of-flight MR angiography was reduced.

central catheter or turbulent flow at the proximal site of the AVF that develops intimal hyperplasia.[1,11] The incidence of IVR following CVD in patients on dialysis remains unclear although numerous reports have been published.[5-9,13] IVR often causes venous congestion that occasionally presents with intracranial hemorrhage or ischemic stroke, mimicking dural AVF.[9] Symptoms of IVR are diverse, including not only nonfocal symptoms such as headache and convulsions but also focal neurological deficits such as double vision and paralysis.[4-7,9,13] Although cases of TN due to arteriovenous malformation or dural AVF have been reported in the literature,[3] it should be noted that IVR due to systemic AV shunt can cause typical Type 1 TN. The imaging features of IVR may be subtle and its true causative site may be distant from the site related to the focal neurological symptoms. IVR following CVD should be considered in the differential diagnosis when imaging findings do not match clinical neurological symptoms in patients on hemodialysis. The true cause may be distantly located from the neurologic structure responsible for the focal neurological symptoms in such patients.

CONCLUSION

Patients on hemodialysis often present with the central venous stenosis or occlusion after the placement of multiple central venous catheters during their long treatment period. We emphasize the importance of considering the involvement of the central venous reflux when local imaging findings are lacking or inconsistent with TN with arterial compression in patients on hemodialysis.

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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