Resolved Cerebral Venous Hypertension after Angioplasty of Central Venous Stenosis in a Hemodialysis Patient: A Case Report

혈액투석 환자에서 발생한 중심 정맥 협착의 혈관성형술 후 호전된 대뇌 정맥 고혈압: 증례 보고

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Stenosis of the central veins is a common complication in hemodialysis patients. However, cerebral venous hypertension and neurological symptoms caused by central vein stenosis are relatively rare. We present a rare case of cerebral venous hypertension in a 63-year-old male who showed venous reflux into the dural sinuses due to central venous stenosis on time-of-flight MR angiography. After management for central venous stenosis, the venous reflux disappeared.

Index terms Intracranial Hypertension; Stenosis; Chronic Kidney Disease; Renal Dialysis; Magnetic Resonance Angiography; Magnetic Resonance Imaging

Received February 18, 2021 Revised April 15, 2021 Accepted April 24, 2021

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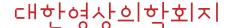
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INTRODUCTION

Hemodialysis patients should have vascular access with adequate blood flow such as from an arteriovenous fistula (AVF) or arteriovenous graft (AVG) to perform hemodialysis. However, a large blood flow volume of vascular access damages vessel walls and can cause stenosis of the central veins. Central vein stenosis often causes venous hypertension resulting in arm and facial edema on the affected side (1). In addition, the rare occurrence of cerebral venous hypertension and cerebral hemorrhage have also been reported (2). However, because jugular venous reflux (JVR) has been regarded as a physiologic phenomenon and other conditions such as cavernous dural AVF can show similar clinical findings (3), it is important to know the imaging findings of cerebral venous hypertension caused by JVR. Here, we report the unusual case of cerebral venous



hypertension caused by severe stenosis at the graft-left internal jugular vein anastomotic site in a hemodialysis patient with an AVG in his left forearm and discuss the imaging features.

CASE REPORT

A 63-year-old male presented with diplopia, dizziness and headache. He was undergoing hemodialysis due to end-stage renal disease caused by hypertension and had undergone about 40 months of maintenance hemodialysis via an AVG in his left forearm. The ophthalmological examination revealed severe left conjunctival congestion. His history and physical examination as well as routine laboratory test values were unremarkable except for renal dysfunction.

Angiography showed severe segmental stenosis of the anastomotic site between the graft and left internal jugular vein. Also there was a backward blood flow from the left internal jugular vein and the flow drained into the right internal jugular vein through the right dural venous sinuses (Fig. 1A). Brain MRI and MR angiography (MRA) showed a time-of-flight (TOF) signal in the intracranial sinuses including the left sigmoid sinus and left cavernous sinus and also the left superior ophthalmic vein (Fig. 1B) without cavernous sinus bulging. A dilated left superior ophthalmic vein was also seen with flow void on T2-weighted imaging (Fig. 1B). No other remarkable findings that could cause the neurological symptoms were seen in the other images. We diagnosed cerebral venous hypertension due to the backward flow of the left internal jugular vein rather than other conditions such as dural AVF.

Percutaneous transluminal angioplasty (PTA) with balloon dilatation and stent placement was performed for stenosis of the graft-left internal jugular vein anastomotic site. The post-stent digital subtraction angiography showed no residual stenosis with mild residual retrograde blood flow along the left internal jugular vein (Fig. 1C). Two weeks after the procedure, the patient's symptoms including diplopia, dizziness, and headache were improved. A follow-up brain MRI was performed three months after the procedure. The TOF signal in the intracranial sinuses and left superior ophthalmic vein had disappeared (Fig. 1D). The patient has been undergoing regular follow-ups at an outpatient clinic without neurologic symptoms.

This study received an ethical review exemption from the Institutional Review Board of author's institution and the requirement for informed consent was waived (IRB No. SCHUH 2021-02-015).

DISCUSSION

Central venous stenosis is a frequent complication in the hemodialysis population and has been an increasing challenge for clinicians including nephrologists and vascular surgeons. About 30% of the hemodialysis population will develop stenosis of the central vein (4), because the high-pressure turbulent flow of hemodialysis stimulates intimal hyperplasia. Due to the high incidence of central vein stenosis, venous hypertension caused by central vein stenosis is a major problem in patients undergoing chronic hemodialysis. The clinical features of central vein stenosis may vary according to the lesion location and in general, the closer the lesion is to the heart, the more severe are the clinical features as in this case. The

Fig. 1. Central venous hypertension due to central venous stenosis in a 63-year-old hemodialysis patient, complaining with diplopia, diziness and headache.

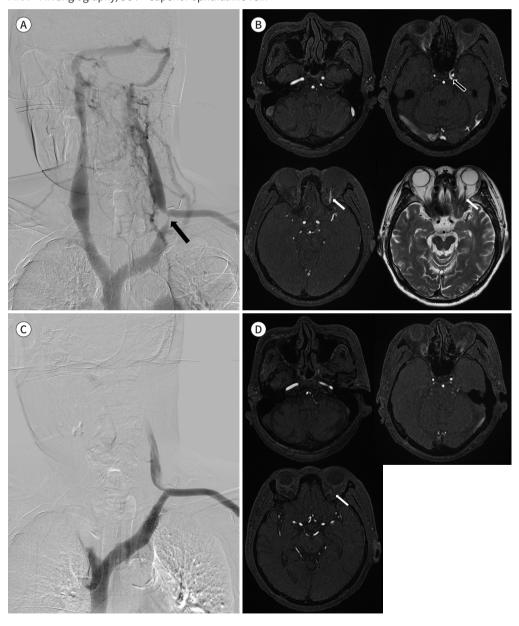
A. Digital subtraction angiography shows tight stenosis between the graft-left internal jugular vein site (arrow), which interferes with antegrade flow and causes retrograde flow into the left internal jugular vein and the intracranial vein. The retrograde venous flow passes through the left sigmoid sinus, transverse sinus, and inferior petrosal sinus and back to the right internal jugular vein.

B. Initial time-of-flight-MRA shows in-flow signals at the intracranial sinuses, including the left sigmoid sinus and the left cavernous sinus (black arrow), correlating with the angiography findings. The left SOV also demonstrates an increased signal (white arrow). T2-weighted imaging also shows dilatation of the left SOV (white arrow).

C. Post-stent digital subtraction angiography shows an improvement in tight stenosis at the graft-left internal jugular vein site with mild residual retrograde flow into the left internal jugular vein.

D. Three months after the procedure, MRA demonstrates no in-flow signals in the venous sinuses nor in the left SOV (arrow).

MRA = MR angiography, SOV = superior ophthalmic vein



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most common clinical manifestation of central vein stenosis in hemodialysis patient is ipsilateral arm and facial edema (1, 2). However, rarely, neurologic symptoms caused by intracranial venous hypertension may also appear. But not all patients who have backflow from the internal jugular vein into the cerebral venous system are symptomatic. If there sufficient venous capacity and interconnection between both transverse venous sinuses, retrograde blood flow may not cause increased cerebral venous pressure. Factors such as disturbance in the blood flow at any point along the collateral circuit or retrograde blood flow beyond the venous capacity may lead to increased cerebral venous pressure and cause neurologic symptoms. Our patient may have initially developed neurologic symptoms from the large amount of retrograde blood flow along the left internal jugular vein due to proximal site high-grade stenosis and the hemodialysis shunt.

Intracranial TOF MRA does not show venous flow, which runs in the craniocaudal direction. Thus, in intracranial TOF MRA images, it is expected that only the arterial structures, which run in the caudocranial direction, will be shown. Therefore, the visualization of venous signals on intracranial TOF MRA can suggest an abnormality. However, JVR can also appear as a physiologic phenomenon on intracranial TOF MRA, which has been reported to range from 1.3% to 6.2% (3). It is clinically important to distinguish between physiologic phenomena and abnormal conditions on MRA. One study of JVR on TOF MRA reported that there are no jugular vein reflux cases with superior ophthalmic vein dilatation (3). Because the superior ophthalmic vein is too far from the central veins for physiologic reflux flow, it is very difficult for the venous reflux flow to reach and dilate the superior ophthalmic vein (5). However, as in our case, severe stenosis of the left internal jugular vein with the hemodialysis shunt can increase the retrograde venous flow, allowing retrograde flow to reach the superior ophthalmic vein and resulting in dilatation. In addition, to differentiate cerebral venous hypertension from cavernous dural AVF with similar clinical features, routine MRI and TOF MRA may be helpful. Because it is rare for retrograde flow to affect the venous flow in cavernous dural AVF cases, the detection of retrograde jugular venous flow on TOF MRA is important to differentiating between JVR and cavernous dural AVF. Also the absence of early opacification of the cavernous sinus in the arterial phase or ipsilateral jugular venous drainage in the arteriovenous phase in contrast-enhanced-MRA was also a reliable finding to suggest JVR (3).

The most common neurological symptom in a patient with cerebral venous hypertension is headache, but various symptoms have been observed in patients with increased cerebral venous pressure. Visual changes, and blindness were reported in a patient with increased intracranial pressure. One study showed visual loss and papilledema caused by occlusion of the brachial-internal jugular graft site and cerebral venous hypertension (6). Another study reported that transient global amnesia (TGA) could be caused by the obstruction of venous drainage (7). This study explains that central venous stenosis could cause venous flow disorders in the deep cerebral veins that drain the hippocampus, resulting in TGA. Also, the persistently increased cerebral venous pressure in the surrounding draining veins can ultimately lead to cerebral infarction or hemorrhage. A case report with cerebral venous hypertension caused by AVG and ipsilateral innominate vein occlusion showed progressive encephalopathy and right occipital infarction (2). And in other cases of intracranial venous hypertension in a hemodialysis patient with AVG, venous intraparenchymal hemorrhages were reported (8, 9).



Endovascular therapy such as PTA is preferred for central vein stenosis because of the surgical limitations due to the presence of lesions in the thorax. And end stage renal disease patients frequently have multiple underlying diseases and are older aged. PTA with balloon dilatation is the primary basis for endovascular therapy and shows a relatively good prognosis. Howevere, when stenosis is severe or recurrent, repeated procedures or stent placement are required.

In our case, the patient was discharged without any neurological sequelae after appropriate procedures including PTA with balloon dilatation and stent insertion. With the appropriate diagnosis and intervention like that our patient received, the symptoms were completely resolved without sequelae.

In conclusion, we reported a case of cerebral venous hypertension by retrograde blood flow from the left internal jugular vein due to central venous stenosis and the associated clinical and radiological findings. Although cerebral venous hypertension by central vein stenosis is rare and shows nonspecific clinical features, through this case report, we emphasize the importance of differentiating it from physiologic jugular reflux or dural AVF. Also through the appropriate diagnostic and therapeutic interventions, severe complications such as cerebral infarction or hemorrhage could be prevented.

Author Contributions

Conceptualizationl, P.S.; data curation, K.H.; investigation, P.S.; project administration, P.S.; resources, P.S.; supervision, P.S.; visualization, all authors; writing—original draft, K.H.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Funding

None

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혈액투석 환자에서 발생한 중심 정맥 협착의 혈관성형술 후 호전된 대뇌 정맥 고혈압: 증례 보고

강희민·박성태*

혈액투석 환자에서 중심정맥 협착은 비교적 흔한 합병증이다. 하지만 중심정맥 협착으로 인해 대뇌 정맥 고혈압과 이로 인한 신경학적 증상이 발생하는 것은 비교적 드물다. 중심정맥 협착으로 인해 발생한 좌측 내경정맥으로부터의 역행성 혈류로 대뇌 정맥 고혈압이 발생한 63세 남성의 증례를 보고하고자 한다. 환자는 중심정맥 협착에 대한 치료를 받은 후 증상 및 치료 전 자기공명영상에서 역행성 혈류를 의미하는 time-of-flight 신호가 소실되었다.

순천향대학교 서울병원 영상의학과