

Case Report

Ruptured vertebrobasilar junction aneurysm unmasking subclavian steal syndrome

Vivek Murumkar, Shumyla Jabeen, Sameer Peer, Aravinda Hanumanthapura Ramalingaiah, Jitender Saini

Department of Neuroimaging and Interventional Radiology, National Institute of Mental Health and Neurosciences, Bengaluru, Karnataka, India.

E-mail: Vivek Murumkar - vivek.murumkar@gmail.com; Shumyla Jabeen - jabeen.shumyla98@gmail.com; *Sameer Peer - sameer.peer602@gmail.com; Aravinda Hanumanthapura Ramalingaiah - aravils@yahoo.co.in; Jitender Saini - jsaini76@gmail.com



*Corresponding author:

Dr. Aravinda Hanumanthapura
Ramalingaiah,
Department of Neuroimaging
and Interventional Radiology,
National Institute of Mental
Health and Neurosciences,
Bengaluru, Karnataka, India.
aravils@yahoo.co.in

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ABSTRACT

Background: Subclavian steal occurs due to stenosis or occlusion of the subclavian artery or innominate artery proximal to the origin of the vertebral artery. Often asymptomatic, the condition may be unmasked due to symptoms of vertebrobasilar insufficiency triggered by strenuous physical exercise involving the affected upper limb. The association of vertebrobasilar junction (VBJ) aneurysms with subclavian steal syndrome has been rarely reported. Hereby, we present a case of VBJ aneurysm associated with subclavian steal treated successfully with endovascular coiling.

Case Description: A 65-year-old female presented in the emergency department with acute severe headache and vomiting with no focal neurological deficits. Non-contrast computed tomography of the brain showed modified Fischer Grade 3 subarachnoid hemorrhage. Subsequent digital subtraction angiogram (DSA) showed VBJ aneurysm directed inferiorly with the left subclavian artery occlusion. There was retrograde filling of the left vertebral artery on right vertebral injection, confirming the diagnosis of subclavian steal. Balloon assisted coiling of the VBJ aneurysm was performed while gaining access through the stenotic left vertebral artery ostium which provided a more favorable hemodynamic stability to the coil mass.

Conclusion: Subclavian steal exerting undue hemodynamic stress on vertebrobasilar circulation can be an etiological factor for the development of the flow-related aneurysms. Access to the VBJ aneurysms may be feasible through the stenosed vertebral artery if angioplasty is performed before the coiling of the aneurysm.

Keywords: Aneurysm, Balloon, Coiling, Stenosis, Vertebral

INTRODUCTION

An intracranial aneurysm develops either due to an underlying defect in the tunica media of a blood vessel or excessive hemodynamic stress exerted on a normal blood vessel.^[12,14] Other factors linked to aneurysm formation and growth are hypertension, smoking, and old age. Posterior circulation aneurysms are less frequent than those of the anterior circulation.^[2] Vertebrobasilar junction (VBJ) is an uncommon location for aneurysm. Fenestration of the basilar artery has been known to be associated with formation of such aneurysm.^[5] Rarely, VBJ aneurysm develops in the setting of subclavian steal syndrome with only few isolated case reports published in the literature.^[1,4] Undue hemodynamic stress exerted on the vertebrobasilar circulation in the presence of subclavian steal is postulated as an etiological factor for the development of such flow related aneurysms.

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CASE DESCRIPTION

A 64-year-old female presented with a history of acute severe headache with vomiting. On examination, she was alert with no focal neurological deficits. Non-enhanced computed tomography (CT) scan of the brain showed modified Fisher Grade 3 subarachnoid hemorrhage (SAH) in the prepontine cistern [Figure 1]. Digital subtraction angiography (DSA) revealed aneurysm of the VBJ measuring 5.6×5.7 mm. There was a small teat projecting downward from the dome of the aneurysm [Figure 1]. On right vertebral angiogram, there was retrograde filling of the left vertebral artery till its origin from the left subclavian artery [Figure 1]. Descending thoracic aortogram showed near-complete occlusion of the left subclavian artery origin [Figure 1]. After multi-disciplinary team discussion, it was decided to proceed with balloon-assisted coiling of the aneurysm. Patient's relatives

were counseled for the procedure, the risks associated with it and informed written consent was obtained for the same.

The procedure was done under general anesthesia. The right femoral artery was punctured with an 18G needle. Arterial access was secured with a 7F long sheath (Rabe Flexor, Cook) placed at the origin of right vertebral artery. 6F guiding catheter (Neuron, Penumbra Inc) was parked in the distal V2 segment of right vertebral artery. A balloon (Eclipse, Balt, France) was parked across the neck of the aneurysm and coiling attempted after cannulating the aneurysm sac with a microcatheter (Headway duo, Microvention, Tustin, CA, USA). However, the coil mass could not be retained inside the sac despite inflation of the balloon due to high flow across the VBJ. It was decided to take access from the left vertebral artery. The stenotic segment of the left subclavian artery was crossed with a 0.035" wire. A 5F vertebral catheter

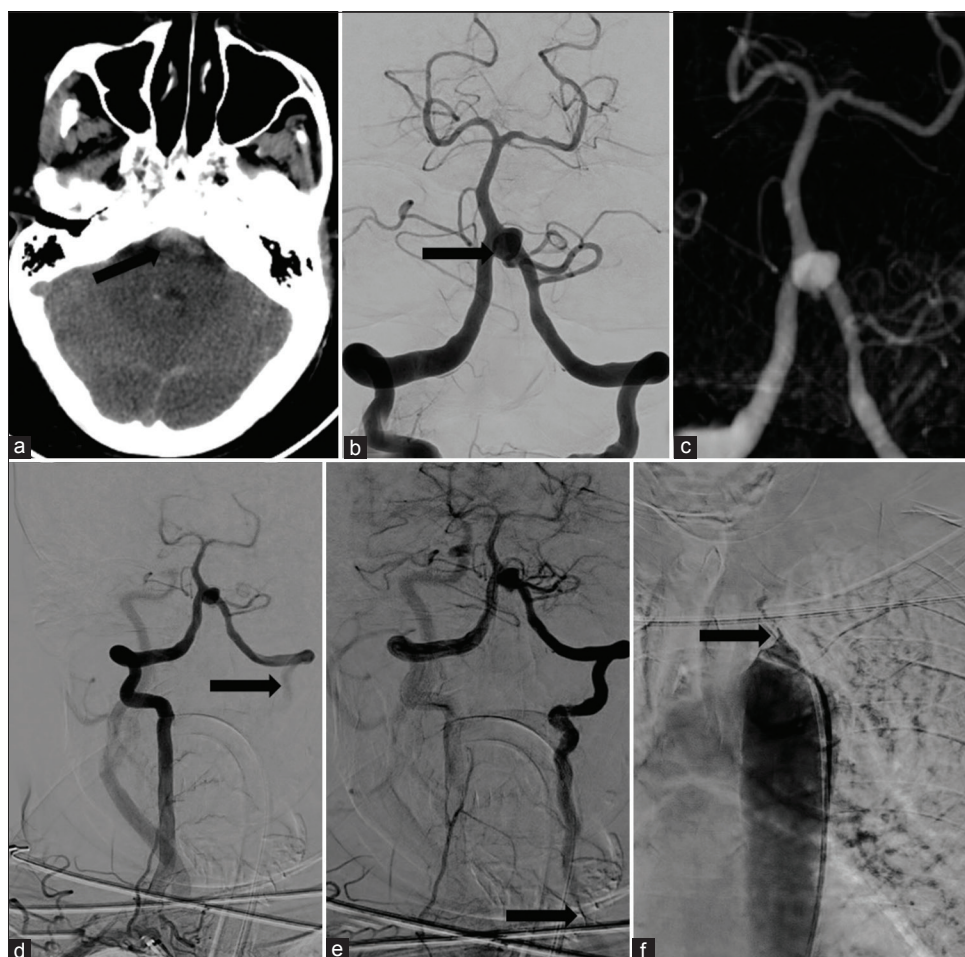


Figure 1: Axial CT brain showing modified Fischer grade 3 SAH in prepontine cistern (black arrow) (a). Right vertebral artery injection showing saccular aneurysm at vertebrobasilar (VB) junction (black arrow) with retrograde flow of contrast in left subclavian (b). 3D rotational angiogram showing saccular aneurysm arising from VB junction. (c) Long sheath injection from the right subclavian artery showing aneurysm in VB junction with retrograde flow in the left vertebral artery (black arrow) (d), next frame of previous run showing progressive retrograde filling of left vertebral artery till its origin from left subclavian artery (black arrow) (e). Aortogram showing tight stenosis of left subclavian origin (black arrow) (f).

was negotiated across the stenosis over the wire and the 7F long sheath was subsequently tracked over it and placed at the origin of the left vertebral artery. The guiding catheter was parked in the V3 segment. Balloon assisted coiling was then successfully performed. Two coils were deployed into the aneurysm sac. Postcoiling angiogram showed Modified Raymond-Roy Class II occlusion of the aneurysm with normal filling of all vessels [Figure 2]. The microcatheter and guiding catheter were withdrawn. Descending thoracic aortogram showed partial opening of the stenotic origin of the left subclavian artery [Figure 2] The 7F long sheath was removed and hemostasis achieved by manual compression. Immediate postprocedure CT showed no new abnormality, no bleed/infarction was noted. Antiplatelets in the form of 150 mg of aspirin were continued for 6 weeks. She had an uneventful hospital stay and was discharged in stable condition.

DISCUSSION

The phenomenon of “Subclavian steal” was first described by Reivich *et al.* in 1961 as a description of two cases and the demonstration of the hemodynamic phenomenon in four dogs.^[9] Fall in blood pressure in the subclavian artery distal to stenosis (below the level of the vertebrobasilar circulation) is considered as an underlying pathophysiological mechanism of subclavian steal syndrome (SSS).^[9] Although often asymptomatic, subclavian steal may manifest in response to hemodynamic stress resulting from increased demand in the upper limb such as vigorous exercise or arteriovenous fistula created for dialysis in the ipsilateral arm.^[4,7] Symptoms range from peripheral ischemia such as arm pain triggered by exercise, paresthesia, cold extremities to vertebrobasilar insufficiency such as drop attacks, dizziness, tinnitus, hearing loss, nystagmus, and diplopia.^[7,8] Rarely, coronary-subclavian steal phenomenon can occur in patients who have undergone coronary artery bypass graft with stenosis proximal to the origin of the internal mammary artery graft used in the procedure.^[11] Signs on physical examination are discrepancy in blood pressure readings between two arms, delayed, and low amplitude pulse on the affected side, bruit in the suboccipital or supraclavicular region.^[7] Diagnosis can be made on color Doppler by the demonstration of flow reversal in the ipsilateral vertebral artery. However, the underlying cause remains elusive which warrants cross-sectional imaging. Phase-contrast magnetic resonance angiogram (MRA) gives information regarding the direction of blood flow while in time of flight MRA, suppression of flow from one direction is performed which can help to diagnose SSS.^[13] DSA is the gold standard for diagnosis and management of SSS.

Intracranial aneurysms associated with SSS are very rare with only handful of cases reported in the literature so far.^[4]

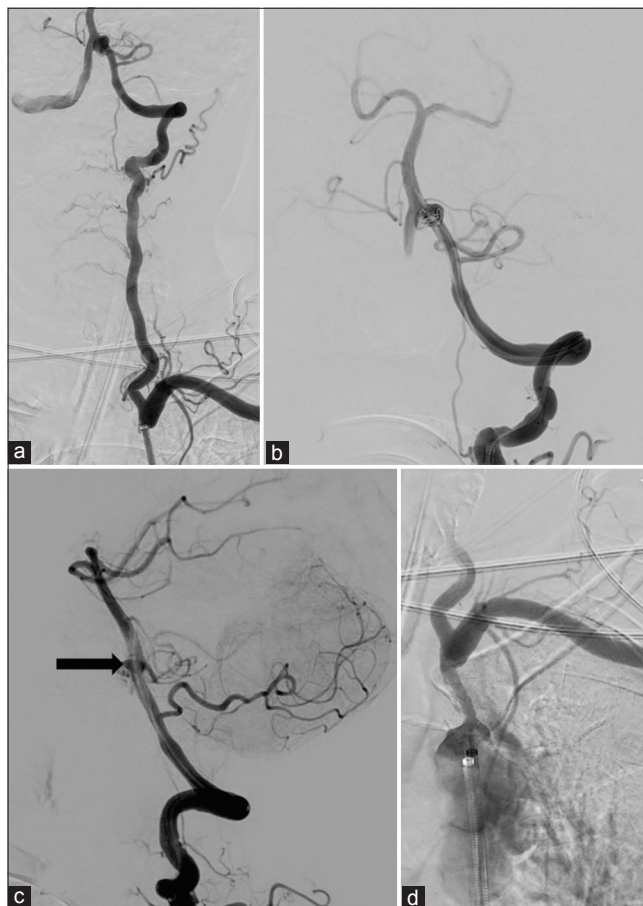


Figure 2: Post angioplasty left subclavian injection showing antegrade flow in the left vertebral artery with filling of aneurysm (a). AP projection of the left vertebral injection after deployment of the first coil in the aneurysm sac (b). Lateral projection of the left vertebral injection after deployment of second coil showing minimal neck residue (black arrow) (c). Post coiling aortogram taken with long sheath in descending thoracic aorta showing residual stenosis at the left subclavian origin (d) note mild opening of the stenosis due to long sheath placement.

Various locations of aneurysms reported in the setting of SSS are VBJ, vertebral artery-posterior inferior cerebellar artery (PICA) junction, posterior communicating artery, and posterior cerebral artery.^[1,4,16] Excessive hemodynamic stress exerted on walls of the posterior circulation arteries due to SSS is postulated as a pathological factor for the development of these aneurysms. The frequent association of VBJ aneurysms with basilar artery fenestration also supports this hypothesis.^[3,6] Treatment of such flow-related aneurysms can be performed by endovascular or microsurgical means. Proximity with the brainstem and abundance of perforators (supplying brainstem and cranial nerve) in the vicinity makes the microsurgical treatment of such aneurysms challenging; hence, the endovascular approach is favored over surgery. Endovascular treatment includes coiling if the neck is narrow and balloon

remodeling, stent-assisted coiling, or placement of flow diverting stents if the neck is wide.^[1,10] Routes for intervention can be from contralateral normal vertebral artery, through ipsilateral vertebral artery after treatment of the stenosis or even through a brachial approach.^[1,4,15,16] In our case, we initially attempted balloon assisted coiling through the contralateral normal side. However, the coil mass protruded into the parent vessel on balloon deflation likely due to increased flow across the VBJ in response to the left subclavian stenosis. Considering risk of inadvertent embolization associated with unstable coil mass, we decided to approach the aneurysm from the left side.

Definitive treatment of subclavian steal syndrome is by endoluminal balloon angioplasty with or without stenting or by carotid-subclavian bypass surgery and axillo-axillary bypass surgery.^[10]

Thus, our case highlights a rare but important association between subclavian steal syndrome and intracranial flow-related aneurysm which can be an initial presenting feature. The exact cause of VBJ aneurysm development in cases of subclavian steal is not known; however, it has been postulated to be a result of hemodynamic stress due to increased flow across the VBJ in case of subclavian steal. This hypothesis is also supported by spontaneous regression of these aneurysms when the flow conditions across the VBJ are restored toward a low flow state. In their case description, Pasco *et al.* demonstrated spontaneous regression in the size of one flow related aneurysm of an intervertebral collateral in case of subclavian steal syndrome after surgical carotid-subclavian transposition.^[5] This case supports the hypothesis that the VBJ aneurysm development in case of SSS is indeed a flow-related phenomenon. Endovascular treatment of such aneurysms is a relatively safe option as compared to surgery and can be performed by navigating through the stenotic side after adequate dilatation of the stenotic segment.

CONCLUSION

Subclavian steal syndrome is often asymptomatic and can rarely be unmasked by the rupture of intracranial flow-related aneurysm. VBJ is the most frequently reported site of this flow-related aneurysm. Endovascular approach is relatively safe for treating such aneurysms which can be performed by gaining vascular access either through the contralateral side or ipsilateral side by performing prior dilatation of the stenotic segment.

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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