

## Case Report

# Ruptured venous aneurysm of cervicomedullary junction

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Received: 18 July 13 Accepted: 11 December 13 Published: 14 January 14

### This article may be cited as:

Aggarwal A, Salunke P, Futane S, Mathuriya SN, Kumar A, Mukherjee KK, et al. Ruptured venous aneurysm of cervicomedullary junction. *Surg Neurol Int* 2014;5:2.

Available FREE in open access from: <http://www.surgicalneurologyint.com/text.asp?2014/5/1/2/124977>

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## Abstract

**Background:** Ruptured venous aneurysm is often seen with arterio-venous malformation (AVM) or developmental venous anomaly (DVA). However, isolated venous aneurysm is unusual.

**Case Description:** We present a case of ruptured venous aneurysm that presented with subarachnoid hemorrhage (SAH) and intraventricular hemorrhage (IVH). Digital subtraction angiography (DSA) revealed a saccular contrast filling pouch in the left lateral aspect of cervicomedullary junction (CMJ). Endovascular intervention was not a viable option. During surgery, a saccular pliable structure approx. 1.5 × 1 cm was found in the subarachnoid space that was clipped and excised. There were no arterial feeders, no evidence of surrounding AVM, and no dilated perimedullary vein.

**Conclusion:** This is perhaps the first reported case of ruptured venous aneurysm (without associated AVM) of CMJ, which was successfully managed surgically. The possible etiologies remain an unnoticed head trauma or a congenital vessel wall abnormality. Surgically clipping and excision remains the treatment of choice for such lesion.

**Key Words:** Arterio-venous malformation, cervico medullary junction, venous aneurysm

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#### Website:

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#### DOI:

10.4103/2152-7806.124977

#### Quick Response Code:



## INTRODUCTION

Venous aneurysm of the cerebral circulation also known as cerebral varix arises mostly in association with arterio-venous malformations (AVM) or with developmental venous anomalies (DVAs) and rarely as singular lesions.<sup>[1]</sup> They rarely present with rupture because the normal venous transmural pressure is low. However, in cases of associated AVM, the high arterial pressure is transmitted to venous side of circulation. This exposes the venous circulation to arterial pressure causing rupture. We present a case of ruptured venous aneurysm present in cervicomedullary junction (CMJ) without

associated AVM or DVA, which was successfully managed by surgery.

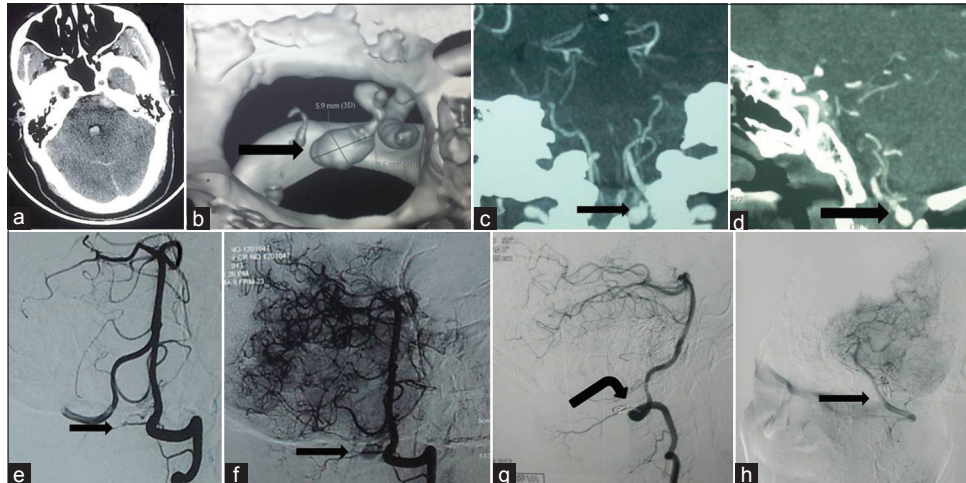
## CASE REPORT

A 52-year-old male with no known previous comorbidities presented with sudden onset headache, vomiting, and transient loss of consciousness. Patient was conscious but disoriented with no focal motor/sensory deficit. Computed tomography (CT) head revealed subarachnoid hemorrhage (SAH) with intraventricular hemorrhage (IVH) [Figure 1a].

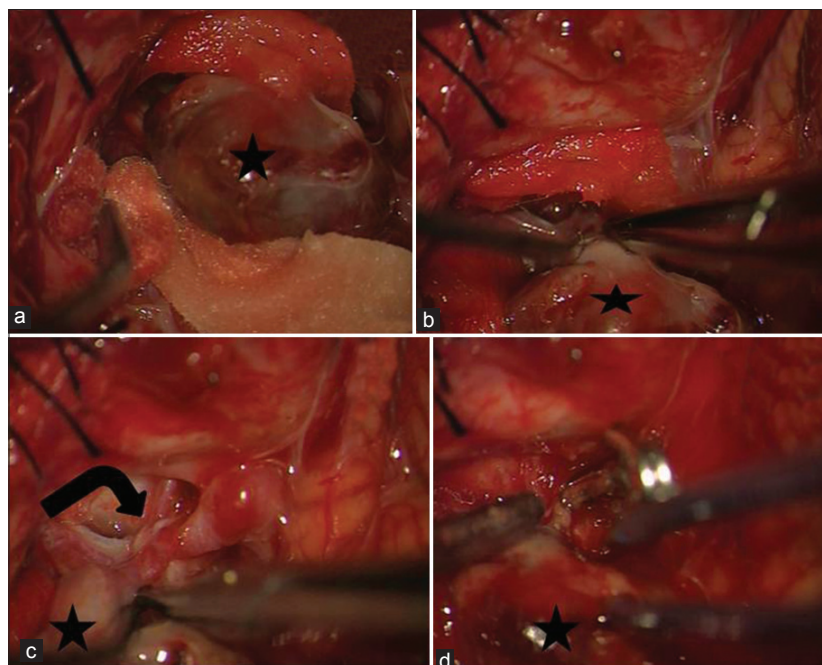
Computerized tomography angiography (CTA) [Figure 1b-d] and digital subtraction angiography (DSA) [Figure 1e and f] revealed a saccular contrast filling pouch in the left lateral aspect of CMJ. The filling was seen in early venous phase on DSA. No arterial feeder could be demonstrated. There was no AVM in the vicinity of the lesion suggesting a diagnosis of solitary alone venous aneurysm.

Patient was operated through a midline suboccipital craniectomy with excision of posterior arch of atlas. Dura

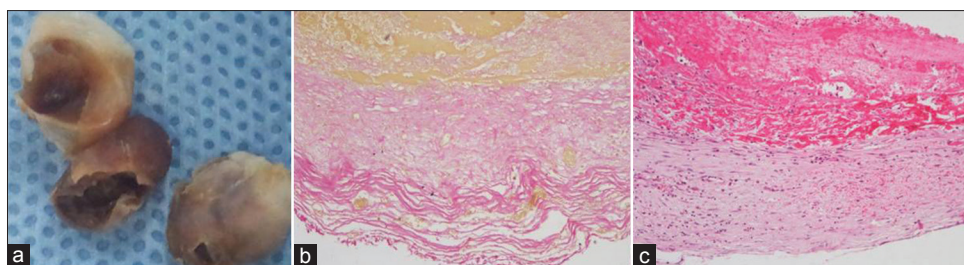
was opened in Y shape. There was no apparent dural AVM. There was evidence of old subarachnoid blood with presence of thick subarachnoid adhesions. A saccular pliable structure approx.  $1.5 \times 1$ cm was found in the subarachnoid space. Its color was indicative of venous blood. Careful dissection revealed the attachment/neck of the aneurysm with a vein. There were no arterial feeders, or evidence of surrounding AVM or dilated perimedullary vein. This aneurysm was clipped and excised [Figure 2a-d]. Gross examination revealed a thin



**Figure 1:** (a) NCCT Head showing SAH and IVH, (b) 3D CT angiography seen from anterior aspect and above showing dilated vascular structure (shown by arrow) near posterior CVJ, (c and d) CT angiography coronal and sagittal reconstruction, showing dilated vascular structure (shown by arrow) near CVJ, (e) DSA in arterial phase does not show any aneurysm, (f) DSA in early venous and late arterial phase showing venous aneurysmal dilatation (arrow), (g and h) postoperative DSA showing nonfilling of aneurysm both in the arterial phase and (g) venous phase, (h) Aneurysm clip is seen



**Figure 2:** Intraoperative images showing: (a) a large pliable vascular lesion (marked with star), (b) dissection being done to identify the neck, (c) the feeding vein (dilated) being skeletonized and (d) aneurysm clip applied at the feeding vein. Aneurysm partially (hidden from view) marked with a star



**Figure 3: (a) Cut section of thin walled Aneurysm with partial thrombus. No arterial ostia was found, (b) Wall of the aneurysm ( $\times 10$ ) in elastic van geison stain does not show elastic fibers and (c) Wall of the aneurysm ( $\times 10$ ) in H and E stain showing absence of elastic fiber layer**

walled vascular structure with no arterial ostia. There was a thrombus present. HPR revealed a single layer of fibromuscular tissue lined [Figure 3a] by a flat endothelium. There were no signs of sclerosis or inflammation. There was no muscle layer [Figure 3b and c]. Patient was discharged on 5<sup>th</sup> postoperative day. Postoperative DSA did not show any aneurysm/AVM. On the last follow up at 5 months after surgery, the patient has no focal deficits and has joined work.

## DISCUSSION

Cerebral venous aneurysms commonly arise in association with AVM or with DVAs and rarely as singular lesions. A case of solitary venous aneurysm left temporo-parietal secondary to head injury has been reported in the past.<sup>[3]</sup> Interestingly, a solitary venous aneurysm at the CMJ has never been reported.

The pathogenesis of cerebral venous aneurysm involves the distention of vein by blood under arterial pressure (as in AVMs). The venous wall being thin is unable to handle the arterial pressure for a long time ultimately producing a weak local dilatation or the so-called venous aneurysm. Another pathogenic mechanism is focal mural injury to venous wall.<sup>[3]</sup>

This case demonstrates that saccular venous aneurysm can arise as a solitary lesion without associated AVM or DVA. Intraoperatively no mass of angiomatous shunt vessel could be identified. Vein was ligated, but there was no dilatation of proximal vessels confirming the absence of AVM.

We were unable to discern the etiology in the present case. There was absence of radiological or intraoperative

AVM. The possibilities remain an unnoticed trauma to head or congenital abnormality of the vessel wall.

The diagnosis rests with DSA. This demonstrates a contrast filling pouch during venous filling phase with or without associated AVM. The management of this entity depends on the presence or absence of AVM. In the absence of AVM, clipping and excision of venous aneurysm offers complete cure as was done in this case.

The present case was unique because of the following reasons. It is perhaps the first documented case of ruptured venous aneurysm without associated AVM at the CMJ in English literature. The treatment of such lesions is open micro neurosurgery. Transarterial endovascular obliteration is difficult because of the tortuosity of arteries in this region. Furthermore, this is not feasible in cases without a demonstrable arterial feeder as in the present case. Transvenous route is not preferable because of the nonavailability of a good venous access to small draining veins and variability of venous anatomy.<sup>[2]</sup> However, during open surgery the venous aneurysm can be safely clipped and excised leading to complete cure. In addition, it provides an opportunity to visualize the involved region for presence of missed pial/dural arterio-venous fistulas.

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