SHORT REPORT

Treatment of Extracranial Vertebral Aneurysm Associated with Two Intracranial Aneurysms—A Case Report

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We report the case of a 42-year-old woman, who was admitted with an acute subarachnoid cerebral haemorrhage, due to the rupture of an end-basilar artery aneurysm. Two other aneurysms were found: one on the sylvian artery and the other on the extracranial vertebral artery. The management of these types of aneurysms is reported as well as a review of extracranial vertebral aneurysm treatment reported in the literature.

Keywords: Vertebral artery; Intracranial aneurysm; Extracranial aneurysm.

Introduction

Intracranial and extracranial vertebro-basilar artery aneurysms can be multiple. Treatment can be endovascular, through open surgery, or both. The authors report treatment of a patient who presented with an extracranial vertebral artery aneurysm associated to two intracranial aneurysms.

Case Report

A 42-year-old woman, who was a heavy smoker and had a medical history of chronic hypertension was admitted to the Emergency Department with a subarachnoid haemorrhage. She complained of a sudden sharp headache earlier that morning, with photophobia, tinnitus and transient motor aphasia. She had no sensory or motor deficit, or loss of consciousness. Her neck was stiff, with diplopia and nystagmus. The CT scan revealed a diffuse meningeal haemorrhage, located in both sylvian fissures and in interhemispheric subarachnoid spaces. A four-vessel angiography was performed, which revealed an extra cranial vertebral aneurysm, associated with two intracranial aneurysms (Fig. 1).

The extra cranial aneurysm was located on the V2 segment of the left vertebral artery, close to the second and the third cervical vertebrae. The aneurysm was sacciform, measuring 11×6 mm with irregular shapes. The first intracranial aneurysm was located at the end of the basilar artery, 8 mm in size with several irregular bulges, and was the suggested cause of the cerebral haemorrhage. The second intracranial aneurysm was located on the M1 part of the right sylvian artery, and had a round regular 6 mm shape. Other vessels (carotid and right vertebral arteries) did not show any other abnormalities; the left vertebral artery was dominant. An anterior spinal artery originated from the V1 segment of the left vertebral artery. The Circle of Willis was intact.

The same day, the end-basilar aneurysm was treated by an endovascular approach; a detachable coil embolization led to a complete aneurysm lumen occlusion. Angiographic follow-up confirmed the positive results of this procedure.

Two weeks later, the right sylvian aneurysm was treated by a surgical pterional approach; the angiogram, 10 days later, confirmed the complete aneurysm exclusion.
One month later, the patient underwent elective surgery for the left extracranial vertebral aneurysm via a upper cervical approach under EEG monitoring; an end-to-end anastomosis was carried out between the occipital artery (branch of the external carotid artery), and the distal V3 segment of the vertebral artery. The V3 proximal segment was ligated. There was no postoperative complication except for a temporary left facial paresis. One month later, the follow-up angiogram confirmed the patency of the bypass and aneurysm occlusion (Fig. 2). The vertebral artery remained patent upstream to the ligation, as the anterior spinal cervical artery originating from the V1 segment of the artery. The facial paresis had almost disappeared.

Discussion

The interest of this case report is the association of extra- and intracranial aneurysms, and their management, particularly for the vertebral aneurysm. Vertebral aneurysms are rare and represent less than 0.5% of all reported aneurysms. They are often located on the intracranial segment of the artery; a series published by Andoh et al. contained 54 vertebral aneurysms among which 10 were located on the extracranial portion. When they are located at this site, they frequently occur on the V1 and V3 segments. However, aneurysms of the V2 segment are extremely rare and are often revealed following complications as rupture, dissection or adjacent organ compression.
Extra cranial vertebral aneurysms rarely occur in atheromatous disease but are more frequently observed in dysplastic pathologies, either in fibromuscular dysplasia, in elastic tissue pathology (Elher-Danlos disease) or in Von Recklinghausen’s neurofibromatosis.3

Multiple aneurysms, with association of intra- and extracranial locations are very uncommon and are rather the subject of isolated reports,4 whereas multiple intracranial aneurysms occur more frequently.

In the case of this patient, a dysplasia was suspected. A sample of vertebral artery was obtained during surgery and was microscopically evaluated but did not confirm the dysplasia. Other aneurysmal localizations were searched, in particular the renal arteries using duplex scan; no aneurysms were found.

In this case, the facial paresis was isolated, without any other neurological deficit. This was due to the compression of the facial nerve by the retractor placed at the distal end of the cervical approach. The facial paresis completely disappeared few weeks later.

Several options to treat this vertebral aneurysm were available; we chose to treat the vertebral aneurysm surgically with immediate re-vascularization of the distal vertebral artery, due to the dominant characteristic of the left vertebral artery. We also chose not to ligate the vertebral artery at the proximal neck of the aneurysm due to the existence of the proximal spinal collateral artery, and to the upper cervical approach, which did not permit a proximal vertebral approach. We expected that the vertebral aneurysm would spontaneously occlude, leaving the spinal artery patent; the follow-up angiogram confirmed this surgical approach. If the aneurysm remained patent after surgery, it was feasible to treat it secondarily using detachable coils or endovascular glue.

Two types of treatment could be proposed for extracranial vertebral aneurysms. The first is surgical treatment, using different methods.

Simple proximal ligation was used by Burger et al. in a V3 vertebral aneurysm.5 This seemingly easy method exposes the patient to the risk of repermeation via the anastomosis between the external carotid branches and the muscular branches of the vertebral artery. Moreover, the ligation of the vertebral artery is not without cerebral ischemic risk and should be preoperatively evaluated by a transcranial Doppler examination. An other method is the proximal and distal ligation of the vertebral artery, at both sides of the aneurysm without revascularization according to Rifkinson-Mann et al.6 This technique entirely excludes the aneurysm, but complications are the same as in the previous method. The third method, as described by Laurian et al., is a proximal and distal ligation associated to a venous bypass which not only permits complete aneurysm exclusion but also maintains the distal artery perfusion.7 This method can be used when a Circle of Willis dysfunction is suspected.

The fourth method, which was used in the present case, is aneurysm exclusion by distal ligation associated to the revascularization of the distal vertebral artery, either via the internal carotid artery, or an external carotid branch. This method, only used in rare situations, was performed in our case due to the existence of a spinal artery upstream to the vertebral aneurysm.

The second type of treatment is endovascular. To date few cases have been reported in the literature,
with no long-term follow-up. Three methods are currently available. An aneurysm embolization can be performed on a small neck aneurysm, by coils or detachable balloon, even on a slightly-mobile portion of the artery. This method should be avoided on the V2 segment where the transverse apophysis can press on the artery and weaken the edge of the aneurysm stiffened by the coils. The association of a stent with placement of detachable coils through the meshes has previously been reported.\textsuperscript{8} This method can be used for an aneurysm with a large neck where embolization alone is not possible; the graft acts as a basic structural support for the detachable coils. Lastly, the use of a stent graft is an alternative which has been recently applied in vertebral artery aneurysms.\textsuperscript{9,10}

We chose to treat this vertebral aneurysm surgically and not via an endovascular approach, due to the localization on the V2 segment of the aneurysm and its wide neck which was, in fact, a contraindication to endovascular treatment. Due to the size (11 mm long) and the irregular shape of the vertebral aneurysm, we were prompted to perform surgical management.

**Conclusion**

Extracranial vertebral artery is an uncommon aneurysmal localization which very rarely concerns the V2 segment. The association of an extracranial vertebral and two intracranial aneurysms has, to our knowledge, never previously been reported. The surgical treatment associating proximal vertebral ligation with re-vascularization of the distal vertebral artery should be considered for a young patient population. Also, the aneurysm should be treated without delay as evolution of dysplastic lesions often has a poor prognosis.

**References**


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