

Case reports

Thrombosed vertebral artery aneurysm presenting with hemorrhage and bulbar compression: report of two cases

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Abstract

Bulbar compression by vertebral artery (VA) aneurysm is extremely rare and only reported in a few cases. We report two cases with thrombosed VA aneurysm compressing medulla oblongata; one presented with bulbar apoplexy hemorrhage and the other with subarachnoid hemorrhage (SAH). A 61-year-old male (case 1) presented with consciousness disturbances, left hemiplegia, and subsequent respiratory arrest. He was subjected to controlled ventilation, and computed tomography (CT) scan showed bulbar hemorrhage. Magnetic resonance imaging (MRI) and digital subtraction angiogram (DSA) revealed fusiform right VA aneurysm with partial thrombosis and bulbar compression. Intra-aneurysmal embolization with Guglielmi detachable coils (GDCs) relieved his clinical symptom including respiratory disturbance. Three months after the onset, he remained moderately disabled. A 76-year-old male (case 2) presented with severe headache and subsequent respiratory disturbance. CT scan on admission showed subarachnoid hemorrhage with acute hydrocephalus. Ventricular drainage rapidly improved consciousness while respiratory disturbance persisted over several days. MRI and DSA suggested spontaneous thrombosis of the right VA dissection with bulbar compression. He showed gradual recovery of his respiration over a week. After ventriculo-peritoneal shunting, he was transferred with moderate disability. These results suggest that the elimination of the pulsatile effect of VA aneurysm adjacent to medulla oblongata can improve symptoms caused by aneurysm-related compression. Early diagnosis and appropriate treatment such as intra-aneurysmal embolization for ameliorating the blood flow inside the aneurysm can relieve mass effect and clinical symptoms.

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1. Introduction

Compression of medulla oblongata can be caused by a variety of vertebral artery (VA) abnormalities such as curved VA, elongated VA, dolichoectatic VA, and VA aneurysm [1–6]. Bulbar compression by VA aneurysm is extremely rare and is only reported in a few cases [6–9]. Furthermore, there is no previous report demonstrating such cases suffering bulbar compression by VA aneurysm following intra-aneurysmal hemorrhage with spontaneous thrombosis or subarachnoid hemorrhage (SAH).

Here, we report two cases with VA aneurysm compressing medulla oblongata, one presented with bulbar apoplexy hemorrhage and the other with SAH.

2. Case reports

2.1. Case 1

A 61-year-old male with past history of hypertension was admitted to our hospital 2 h after a sudden onset of severe vertigo, weakness of the left limbs, and inarticulation. He was semi-comatose on admission, and presented left hemiplegia. During physical examination, he presented sudden respiratory arrest and was endotracheally intubated and ventilated. The computed tomography (CT) scan demonstrated

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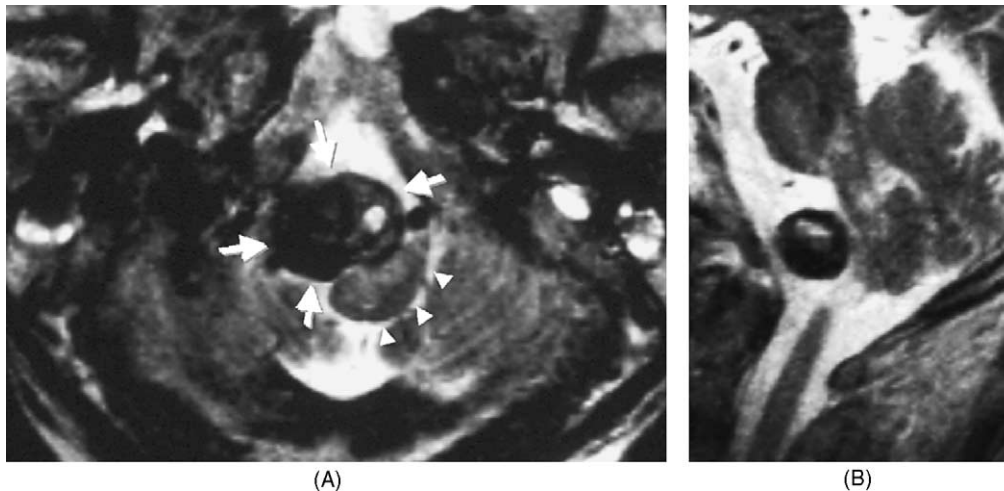


Fig. 1. Axial (A) and sagittal (B) magnetic resonance imaging (MRI) demonstrating high intensity lesion associated with low intensity lesion on T2-weighted image (T2WI), suggesting partial thrombosed vertebral artery (VA) aneurysm (arrows), which compresses medulla from right anterior side (arrow heads) (A). High intensity lesion on T2WI, suggesting edema, is also shown in compressed medulla (B).

bulbar apoplexy hemorrhage without SAH (data not shown). We initially considered that his neurological deficit was due to hypertensive bulbar hemorrhage, thus we treated him conservatively under blood pressure control with mechanical ventilation for a month after tracheostomy. Magnetic resonance (MR) imaging showed heterogeneous mass lesion associated with low intensity signal rim, 20 mm in diameter, compressing medulla oblongata from right anterior side (Fig. 1A). Medulla oblongata adjacent to the mass lesion showed high intensity on T2-weighted images (T2WI), suggesting edema formation due to compression by hematoma (Fig. 1B). Digital subtraction angiogram (DSA) demonstrated fusiform right VA aneurysm located at the distal side of the origin of ipsilateral posterior inferior cerebellar

artery (PICA) (Fig. 2A and B). Based on these findings, we finally diagnosed partial thrombosed fusiform aneurysm of right VA with hemorrhage. This patient was treated by intra-aneurysmal embolization with Guglielmi detachable coils (GDCs) with parent artery occlusion 4 weeks after onset. Post-embolization follow-up angiograms demonstrated complete obliteration of the aneurysm and preservation of its ipsilateral PICA (Fig. 3A and B). Endovascular obliteration of the aneurysm relieved his respiratory disturbance, and he received continuous positive air pressure over 7 days. One month after the embolization, he was completely free from respiratory device. Three months after the onset, he was moderately disabled, and was transferred to another hospital for rehabilitation.

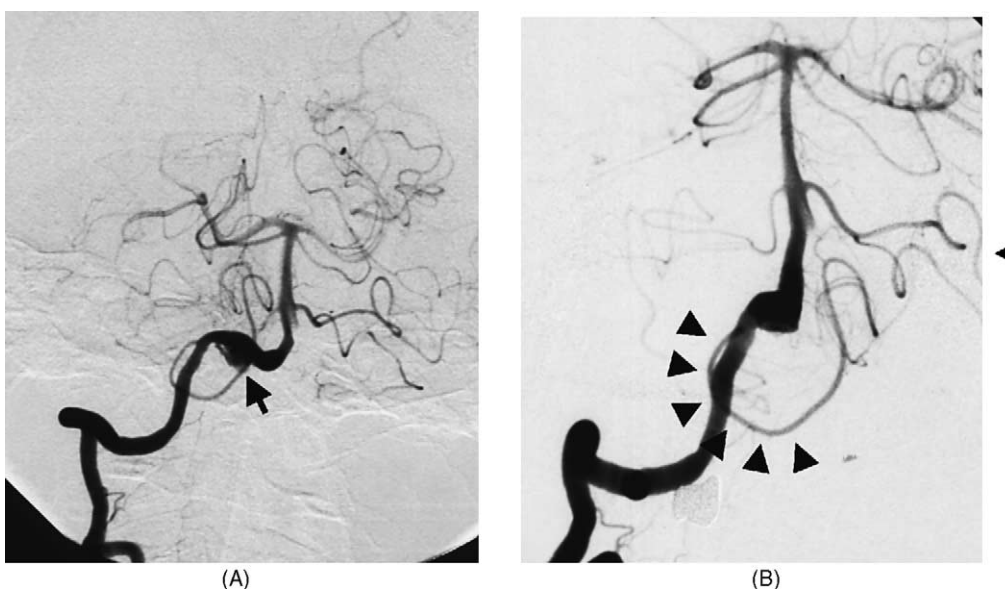


Fig. 2. Anteroposterior (A) and left anterior oblique (B) view of the right VA angiograms demonstrating fusiform aneurysm (arrow) distally to the posterior inferior cerebellar artery (PICA) (arrow heads).

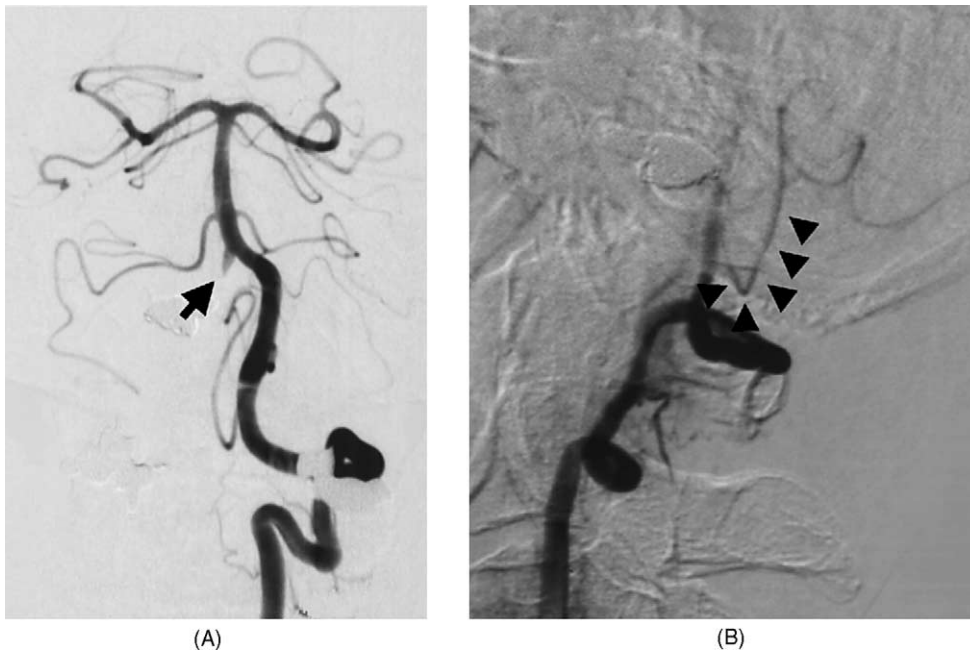


Fig. 3. Post-embolization anteroposterior (A) and lateral (B) view of the bilateral VA angiograms demonstrating occlusion of aneurysm (arrow) (A). PICA is intact (arrowheads) (B).

2.2. Case 2

A 76-year-old male was admitted to our hospital 2 h after a sudden onset of severe headache. He was disoriented on admission, and CT scan demonstrated SAH, associated with acute hydrocephalus, in the interpeduncular fossa, the bilateral ambient cisterns, third and fourth ventricles (Fig. 4). His consciousness and respiratory function deteriorated on the day of admission, and he was endotracheally intubated and received controlled ventilation. Emergency ventricular

drainage improved his consciousness, while his respiratory dysfunction persisted over several days. He showed gradual recovery of his respiratory dysfunction over a week. MR angiography suggested right VA occlusion proximal to the VA union, while left VA and basilar artery were intact (data not shown). Digital subtraction angiogram 6 days after the onset demonstrated right VA occlusion and retention of the contrast material at the distal side of the occlusion (Fig. 5A), suggesting spontaneous thrombosis of the right VA dissection. Left VA angiogram revealed that left VA and basilar

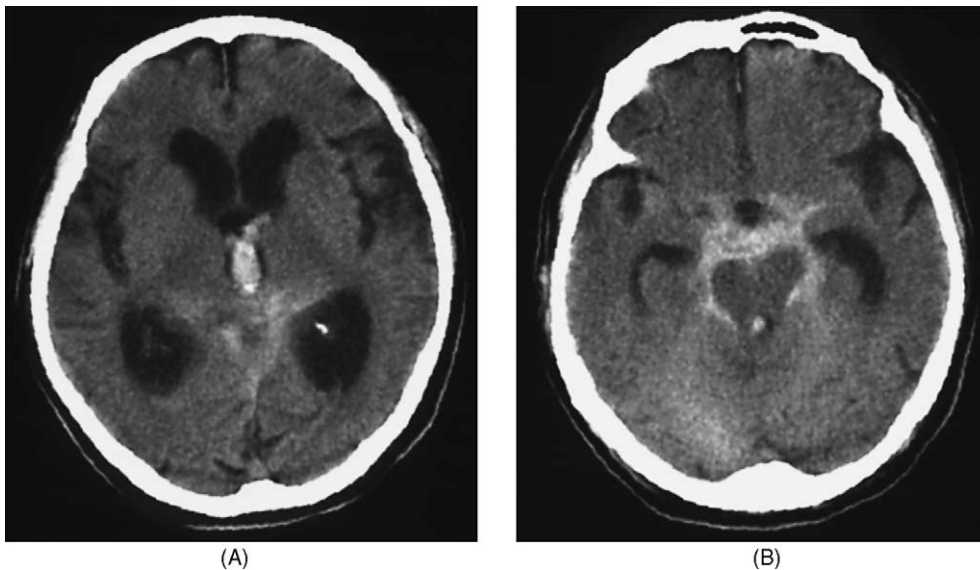


Fig. 4. CT scan on admission demonstrating SAH, associated with acute hydrocephalus (A), in the interpeduncular fossa, the bilateral ambient cisterns, third and fourth ventricles (B).

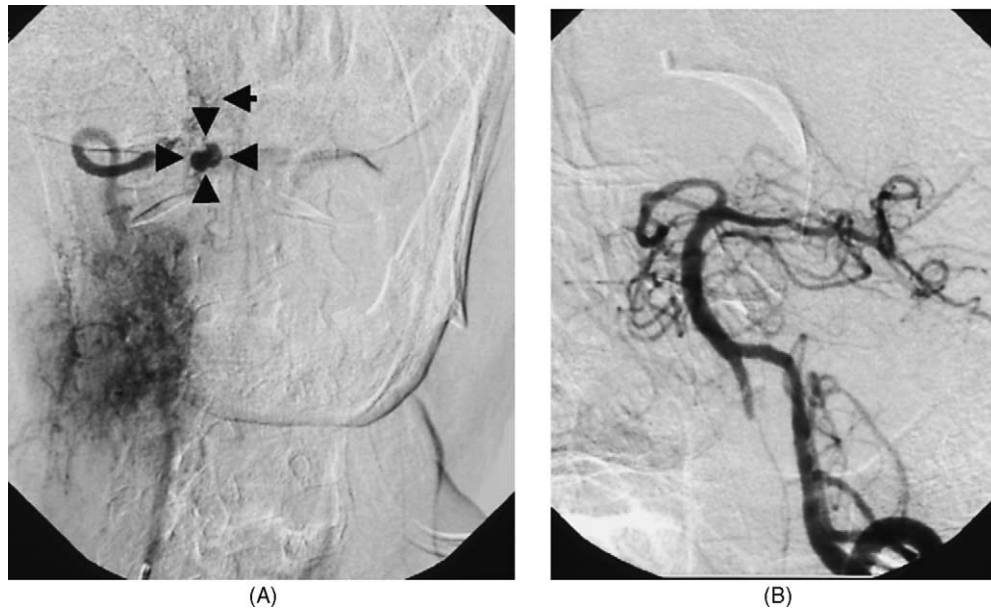


Fig. 5. Anteroposterior view of the right VA angiogram demonstrating VA occlusion just below the foramen magnum (arrow) and retention of the contrast material just on medial side of the occlusion (arrow head) (A). Left anterior oblique view of the left vertebral angiogram demonstrates retrograde filling to the distal end of right VA, while basilar artery and left VA are not affected (B).

artery were not affected (Fig. 5B). The MR imaging revealed mass lesion associated with low intensity signal rim, 5 mm in diameter, compressing medulla oblongata from right anterior side (Fig. 6A). Medulla oblongata showed high intensity by T2WI, which indicated edema formation due to compression by thrombosed aneurysm (Fig. 6B). We carefully followed-up this patient under strict blood pressure controlled, and he did not suffer re-rupture of the aneurysm or cerebral vasospasm. Follow-up angiogram 4 weeks later showed significant decrease of the contrast retention (data not shown). The ventriculo-peritoneal shunt was performed 5 weeks after the onset, and he was transferred to other hospital with moderate disability.

3. Discussion

Compression of the medulla oblongata can cause a variety of signs and symptoms such as hemiparesis, quadriplegia, lower cranial nerve deficits, myoclonus, respiratory disturbance including central sleep apnea, and downbeat nystagmus [1,2,5,6,10,11]. A case with bulbar compression by unruptured giant VA aneurysm presenting with respiratory disturbance has been reported previously, who was successfully treated by surgical removal of aneurysm [6]. Similarly, surgical removal of the mass lesion or neurovascular decompression has been reported to improve such signs and symptoms [1,2–4,6,12–14]. A patient with

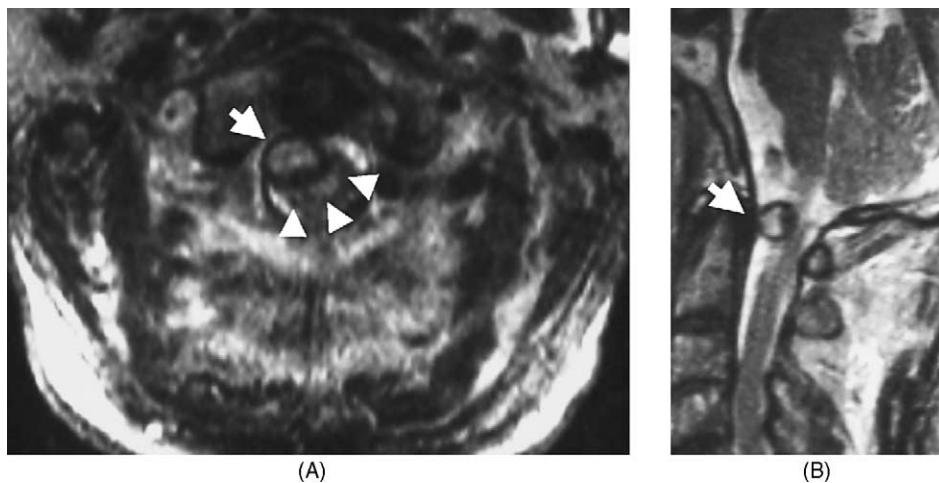


Fig. 6. Axial (A) and sagittal (B) MRI demonstrating high intensity lesion associated with low intensity signal rim on T2WI (arrow), which compresses medulla from right anterior side (A and B). High intensity lesion on T2WI is detected in medulla (arrow head) (A).

brain stem syndrome secondary to VA dolichoectasia was successfully treated by neurovascular decompression [5]. Similarly, another case with pyramidal tract signs by bulbar compression was successfully treated by VA resection after the failure of neurovascular decompression [2]. Regarding VA aneurysm compressing adjacent structure, direct surgical treatment including resection with or without anastomosis has been considered to be optimal although there is a difficulty in its proximity to highly vulnerable neural structures such as the brain stem and cranial nerves. However, it is still undetermined whether intra-aneurysmal embolization without aneurysmal resection can improve signs and symptoms caused by bulbar compression. In our series, the first case showed significant improvement of his respiratory disturbance after the intra-aneurysmal embolization with parent artery occlusion of fusiform VA aneurysm compressing medulla oblongata. Similarly, the second case showed spontaneous thrombosis of the VA aneurysm after SAH and subsequent recovery of his respiratory dysfunction. These results suggest that the elimination of the pulsatile effect of VA aneurysm on the medulla oblongata can improve symptoms caused by the bulbar compression by the aneurysm. In fact, disappearance of the facial spasm due to aneurysmal compression was reported following intra-aneurysmal embolization of the VA aneurysm [9].

The growth mechanism of the thrombosed aneurysm is controversial. Nagahiro et al. [15] suggested that development of the intra-thrombotic capillary channels might be an important factor. Alternatively, simple dissection of the aneurysmal wall through many clefts in the old thrombus might be involved in the growth and rupture of thrombosed aneurysms of the VA [16]. However, rapid enlargement of the VA aneurysm following SAH or intra-aneurysmal hemorrhage and subsequent manifestation of the respiratory disturbance due to bulbar compression is uncommon. In our series, case 1 presented intra-aneurysmal hemorrhage and rapid expansion of the aneurysm, which may have caused the compression of the medulla oblongata. Case 2 presented with SAH due to VA dissection, and MR angiography and VA angiogram suggested spontaneous thrombosis at the acute stage. Thrombus formation at the false lumen may have resulted in parent artery occlusion as well as in the formation of the mass lesion causing the bulbar compression. Since aneurysm can even grow after complete thrombosis [6,8], careful follow-up is necessary in our series.

In summary, we reported two cases with thrombosed VA aneurysm compressing medulla oblongata; one presented with bulbar hemorrhage and the other with SAH. Rapid enlargement of the aneurysm following hemorrhage may cause respiratory disturbance due to bulbar compression.

Early diagnosis and appropriate treatment such as intra-aneurysmal embolization for ameliorating the blood flow inside the aneurysm can relieve this symptom.

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