



Vertical Bow Hunter's Syndromes Caused by C1 Pedicle Screw Compression: Case Report and Literature Review

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Abstract

Bow Hunter's syndromes are well-described reproducible Vertebral Artery (VA) compression syndromes that occur during horizontal head rotation. It's rare that occurred during neck extension and caused by C1 pedicle screw compression, which was so-called vertical Bow Hunter's syndromes. Here, an adult patient was reported who received C1-C2 pedicle screws and plates fusion surgery and suffered from dizziness when extending his neck after one year postoperatively. Repeated basilar artery occlusions were also noted. Dynamic angiography confirmed left VA stenosis caused by C1 pedicle screw compression during neck extension. As we know, this is the first iatrogenic case of vertical Bow Hunter's syndromes. After screw removal, left VA pseudoaneurysm and cerebellar infarction with hemorrhagic transformation were found unexpectedly. Endovascular placement of covered stent was used for obliteration of the pseudoaneurysm. The patient had good clinical recovery without residual sequela after treatment.

Keywords: Bow Hunter's syndromes; Vertebral artery compression; C1 pedicle screw; Vertebral artery pseudoaneurysm

Background

Bow Hunter's syndromes are well-described reproducible Vertebral Artery (VA) compression syndromes that occur during horizontal head rotation. The mechanical occlusion of the VA which causes a reduction in blood flow in posterior cerebral circulation results in transient reversible symptomatic vertebrobasilar insufficiency [1-3]. It's rare that occurred during neck extension. Here, we presented an adult patient with VA compression syndromes during neck extension which caused by C1 pedicle screw compression. This type of VA compression syndromes occurred during neck extension, which was so-called "vertical Bow Hunter's syndrome". As far as we know, this vertical type of Bow Hunter's syndromes was reported for the first time.

Case Presentation

This adult patient who had underlying diseases of hypertension, dyslipidemia and benign prostate hyperplasia complained of neck stiffness and bilateral hands numbness for years. Cervical spine flexion and extension X-rays showed C1-C2 subluxation. Cervical spine MRI showed C1 cord compression. The patient received decompression surgery of C1 and upper C2 laminectomy; fixation surgery with C1-C2 pedicle screws plus plating and fusion with autologous bone graft plus Actifuse[®] bone graft substitute through posterior approach (Figure 1A, 1B). The patient's symptoms of myelopathy were much improved after operation. About one year later, the patient complained of intermittent dizziness and the symptoms were exaggerated during neck extension. Then, the patient presented to Emergent Room (ER) with comatose twice within 6 months. Repeated BAOs were confirmed by brain CTA and the patient received urgent Intra-Arterial (IA) thrombectomy two times. The acute symptoms were relieved after IA procedure. However, daily activities become struggling. The patient suffered from dizziness as long as extending the patient's neck. Therefore, the patient had no choice but kept neck flexion. Furthermore, the patient could only sleep while sitting up. Due to repeated BAOs and young strokes, Dynamic Angiography (DSA) was arranged for survey. Dynamic angiography showed C1 screw related left VA stenosis with diameter variation. The diameter of VA was 2.3 mm during neck flexion and 1.6 mm during neck extension. The 3D reconstructive images showed dynamic external compression of left VA by C1 pedicle screw (Figure 2). CT of cervical spine also confirmed left C1 posterior arch superior wall breach with screw exposure

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Figure 1: Cervical spine CT in bone window. Sagittal and coronal view of immediately postoperative image (A, B) showed appropriate instrumentation. Comparing with the image one year after operation (C, D), which revealed left C1 posterior arch breach at superior wall with screw exposure and dynamic VA compromise.

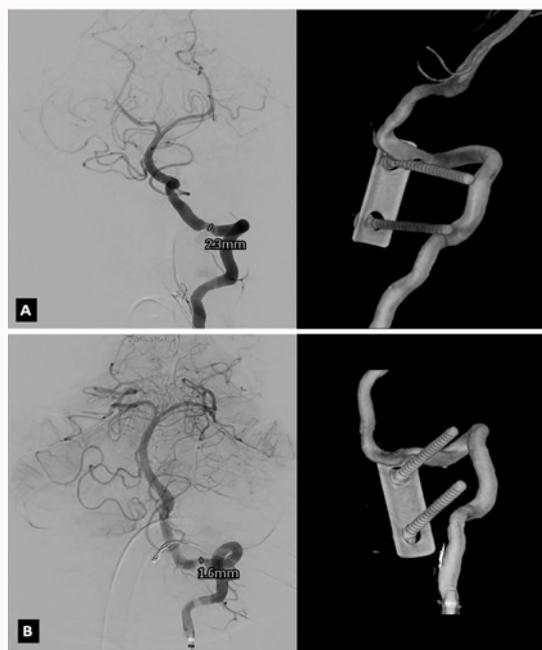


Figure 2: Dynamic DSA of left VA with 3D reconstruction. Diameter of VA was 2.3 mm when neck flexion (A), and decreased to 1.6 mm when neck extension (B). The 3D reconstructive images showed dynamic external compression of left VA by C1 pedicle screw.

results in dynamic VA compromise at sulcus arteriosus (Figure 1C, 1D). Therefore, removal of C1-C2 screw and plate were performed. No active VA bleeding was noted after screw removal. Well fusion and stability were confirmed intraoperatively. However, the patient still felt dizzy postoperatively. Cervical spine Magnetic Resonance Image (MRI) was arranged and showed a pseudoaneurysm at V3 segment of left VA, just at the sulcus arteriosus (Figure 3A). Acute infarction at left PICA territory with hemorrhagic transformation was also noted (Figure 3B, 3C). The infarction may be related to thrombus from the pseudoaneurysm. We used endovascular treatment with covered stent for VA pseudoaneurysm. Preoperative DSA showed

patent posterior circulations and confirmed left VA pseudoaneurysm formation without obvious contrast extravasation. (Figure 4A) PK Papyrus covered stent 3.5 mm × 20mm (Biotronik, AG, Bulach, Switzerland) was deployed under Trojan horse technique (Figure 4B, 4C). Obliteration of pseudoaneurysm was completed (Figure 4D). The patient felt much improved and was discharged smoothly. After 2 weeks, the patient could do a full neck extension without any symptom at the follow-up neurosurgery outpatient department.

Discussion

Reproducible VA compression syndrome was first published by Tatlow and Bammer in 1957 [1]. They hypothesized that symptomatic head rotation may be due to mechanical compression of the VA and observed compressions at C1–C2 and C5–C6. In 1978, the term Bow Hunter's syndrome was first proposed by Sorensen that an archer who presented posterior circulation symptoms when shooting with horizontal head rotation [2]. The mechanical occlusion of the VA which leads to a reduction in blood flow in posterior cerebral circulation resulting in transient reversible symptomatic vertebral basilar insufficiency. Most of the Bow Hunter's syndromes were caused by bony abnormality and occlusion/insufficiency during horizontal head rotation. This case, however, presented Bow Hunter's syndromes during neck extension (vertical type) caused by trauma which was quite rare. There were two case reports of VA compression syndromes during neck extension. Safin et al. [4] reported a case that had multiple posterior fossa strokes and was found to have dynamic occlusion of right VA visualized via cerebral angiogram upon extension of her neck. Yun et al. [5] reported a case presented recurrent vertigo occurred only during neck extension, and resolved immediately on resuming the neutral head position. These two cases had no trauma history, and VA compression was caused by cervical spondylosis or congenital fusion with instability. Posterior atlantoaxial (C1-C2) fusion surgery was used to treat atlantoaxial instability. Since the beginning of the 20th century, various fusion techniques have been developed with improved safety profile, higher fusion rates, and superior clinical outcome. Wiring, hooks, transarticular screw and lateral mass screw fixation including Goel technique and Harms technique [6]. In this case, we used modified technique with



Figure 3: Cervical spine MRI/MRA. MRA (A) demonstrated a pseudoaneurysm at V3 segment of left VA, just at sulcus arteriosus which was injured after C1 lateral mass screw removal. Sagittal and coronal view of T2 weighted image (B, C) showed acute infarction at left PICA territory with hemorrhagic transformation resulted from pseudoaneurysm.

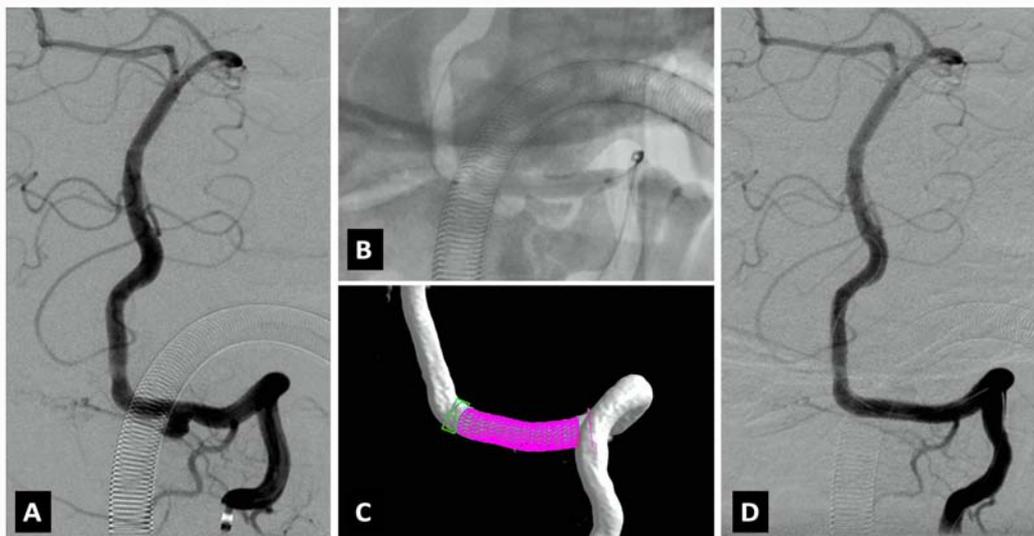


Figure 4: Endovascular treatment of pseudoaneurysm. DSA of left VA (A) confirmed pseudoaneurysm formation. After deployment of a balloon-mounted PK Papyrus covered stent 3.5 mm × 20 mm (B, C), complete obliteration of pseudoaneurysm was achieved immediately (D).

transpedicular screw/plate fixation which was proposed by Chen JF, Wu CT et al. in 2005 [7]. During the procedure, there is no need to sacrifice the bilateral C-2 ganglion or to expose the C-2 isthmus. Thus, we can shorten the operative time, prevent troublesome venous bleeding, and decrease blood loss during the procedure. This technique is simple and easy to perform and can lessen the risk of vessel and/or neural damage. Current case of VA compression syndrome was caused by C1 pedicle screw compression and possibly due to superior wall breach. Especially, the patient presented delayed symptoms about one year after C1-C2 fusion surgery. Complications of posterior approach of screw fixation were reported when atlas in the VA groove (C1 pedicle) was ≤ 3.5 mm in height [8]. In the case series, superior and inferior walls of the pedicles were found to have certain degree of damage respectively and total wall breakage rate was 36.4%. The atlas pedicle screws could be properly extruded, which might result in a risk of damage to the VA. Although there

was no active VA bleeding after screw removal, pseudoaneurysm and cerebellar infarction with hemorrhagic transformation were found unexpectedly. This acute embolic cerebrovascular accident localized to PICA territory as determined by MRI, manifest dizziness. Compared to previous image study, including brain CTA and DSA, the most plausible source of this embolic stroke was ascribed to the VA pseudoaneurysm, and thromboses could be pumped out during neck flexion and extension. Because of the perceived risk of further embolic stroke and risk of rupture, surgical management of pseudoaneurysm was recommended. Surgical management of VA injuries is technically difficult because of the critical location. Traditionally, most of these injuries are either observed, ligated surgically, or embolized [9]. Recently, cover stent become a treatment option in terms of time and invasiveness, as it provides immediate sealing of the lesion and allows salvation of the VA. In the proper scenarios, deployment of covered stents can lead to exclusion of pseudoaneurysms with conservation

of patency of the vessel [10]. Therefore, follow-up DSA or intraoperative angiography after removal of C1 screw is required for the risk of VA injuries.

Conclusion

Intraoperative vascular injuries in cervical spine surgeries were rare, but carried significant morbidity and mortality when they did occur. We reported this rare case with vertebral artery compression syndromes during neck extension caused by left vertebral artery stenosis compressed by C1 pedicle screw. It was the so-called "Vertical Bow Hunter's syndromes". Dynamic angiography was recommended for diagnosis. On the other hand, endovascular placement of covered stent was a safe and effective treatment option for symptomatic vertebral artery pseudoaneurysm.

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