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Spontaneous atraumatic vertebral artery occlusion due to physiological cervical extension

Case report

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Vertebral artery (VA) occlusion is a serious and potentially life-threatening occurrence. Bow hunter's syndrome, a mechanical occlusion of the VA due to physiological head rotation, has been well described in the medical literature. However, mechanical VA compression due to routine flexion or extension of the neck has not been previously reported. The authors present the unique case of a woman without any history of trauma who had multiple posterior fossa strokes and was found to have dynamic occlusion of her right VA visualized via cerebral angiogram upon extension of her neck. This occlusion was attributed to instability at the occipitocervical junction in a patient with a previously unknown congenital fusion of both the occiput to C-1 and C-2 to C-3. An occiput to C-3 fusion was performed to stabilize her cervical spine and minimize the dynamic vascular compression. A postoperative angiogram showed no evidence of restricted flow with flexion or extension of the neck. This case emphasizes the importance of considering symptoms of vertebrobasilar insufficiency as a result of physiological head movement. The authors also review the literature on VA compression resulting from physiological head movement as well as strategies for clinical diagnosis and treatment.
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Key Words • **vertebral artery** • **occlusion** • **extension** • **stroke** • **cervical**

Vertebral artery (VA) occlusion is a serious, morbid, and potentially life-threatening occurrence. It can result in devastating strokes within the brainstem and/or cerebellum, causing transient or permanent neurological deficits, and in some instances coma or death. Insufficiency of the vertebrobasilar system can present with symptoms of diplopia, dysarthria, visual disturbances, vertigo, sensory or motor symptoms, or syncope. The etiology leading to VA occlusion is typically atherosclerotic, thromboembolic, traumatic, or mechanical in nature.

Mechanical VA compression has been reported with chiropractic manipulation,[3,9,15,23] surgical positioning,[25,34] rheumatoid subluxation,[11,16,17,20] and a multitude of physical and sport activities.[8,29,30,35] Bow hunter's syndrome, a mechanical occlusion of the VA due to physiological head rotation, was first described in 1978 in an archer who presented with posterior fossa symptoms.[32] Since then, multiple other cases have described mechanical VA stenosis or occlusion during routine head rotation.[26–28] Although the entity of rotational VA occlusion due to physiological head rotation has been well described and reviewed,[5,7,18,19] there currently exists no literature on this entity occurring with either physiological flexion or extension of the neck. We present a unique case of a woman without any history of trauma, who had multiple posterior fossa strokes with extension of her neck and was found to have dynamic occlusion of her right VA. We also review the literature on VA compression due to physiological head movement and management strategies for both clinical diagnosis and treatment.

Abbreviations used in this paper: VA = vertebral artery; VBI = vertebrobasilar insufficiency.

Case Report

History and Examination. A 37-year-old woman with no medical history was transferred to Tufts Medical Center from an outside hospital for evaluation of stroke. One month prior she had presented to the outside institution with reports of vertigo, tightness in the right occipital region of her head, as well as progressive headaches that were associated with dimming of her entire visual fields.

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On presentation, she was neurologically intact. A CT angiogram was obtained, demonstrating a right cerebellar infarct (Fig. 1A). A hypercoagulable panel, as well as both transthoracic and transesophageal echocardiograms, were performed and were normal. She was started on 81 mg of aspirin and had Holter monitoring that did not show any cardiac arrhythmias. A congenital anomaly of the craniocervical junction was not appreciated at this time. She returned to the same institution a month later with sudden onset vertigo associated with nausea, tightness now in the left occipital region of her head, as well as a progressive onset of headache. A second CT scan as well as MRI demonstrated a new left cerebellar infarct (Fig. 1B–D). She was transferred to our institution for further evaluation of these recurrent strokes. When reviewing her radiological studies, a congenital fusion of the occiput to C-1 as well as a congenital fusion of C-2 and C-3 were appreciated (Fig. 2A and B). Dynamic flexion-extension radiographs were obtained and demonstrated translation of the occiput–C1 fusion mass in relation to the C2–3 fusion mass (Fig. 2C and D).

Patient Management. It was postulated that there was likely VA compromise upon dynamic flexion and extension of the patient's neck. The patient was maintained in a rigid cervical collar. A diagnostic cerebral angiogram was performed and included VA angiograms in the neutral, flexed, and extended positions (Fig. 3). Complete occlusion of the right VA was noted in slight extension with normal flow in other positions (Fig. 3).

Operative Procedure. The patient was recommended to undergo an occiput to C-3 fusion to stabilize her cervical spine as well as minimize the dynamic vascular compression. She had her collar removed and was held in inline traction in slight flexion during intubation. Her collar was then replaced and baseline neuromonitoring was performed with normal motor and sensory evoked potentials. She was placed in 3-point Mayfield pin fixation and turned prone onto a Wilson frame. She was positioned slightly flexed and intraoperative fluoroscopic guidance was obtained. We compared our fluoroscopic films to the preoperative angiographic films and assessed the angles between the anterior skull base, clivus, and temporal floor in relation to the base of C-2 to ensure an appropriate amount of flexion. She underwent occipital plate and C2–3 translaminar screw placement given her anomalous anatomy. Instrumentation placement was confirmed with intraoperative fluoroscopic guidance. There were no changes in motor or sensory evoked potentials. Postoperatively the patient was brought for a CT angiogram to confirm flow within both VAs.

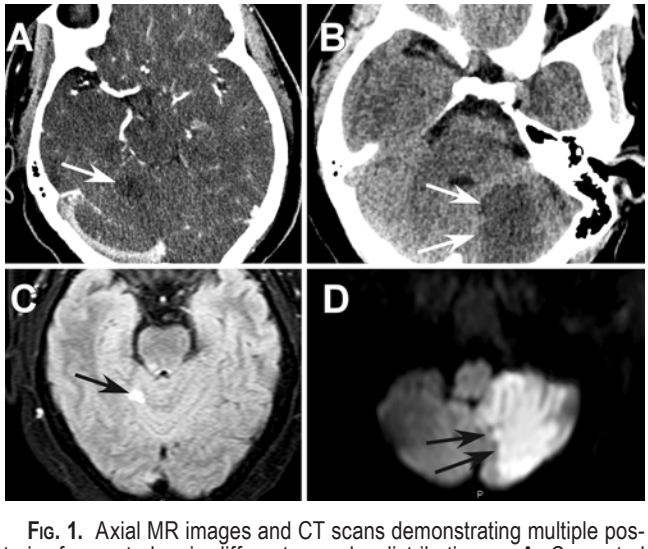


Fig. 1. Axial MR images and CT scans demonstrating multiple posterior fossa strokes in different vascular distributions. **A:** Computed tomography angiogram of the head at initial presentation. The *arrow* highlights a hypodense area in the right cerebellum consistent with stroke. **B:** Head CT scan obtained on second presentation, 1 month after initial presentation. The *arrows* indicate a large hypodense left cerebellar stroke. **C:** Magnetic resonance imaging FLAIR sequence of the head obtained at second presentation. The *arrow* indicates evolving stroke in the right cerebellar hemisphere. FLAIR signal is also noted within the dorsal brainstem. **D:** Diffusion weighted MRI sequence of the head obtained at second presentation. The *arrows* highlight an acute left cerebellar stroke.

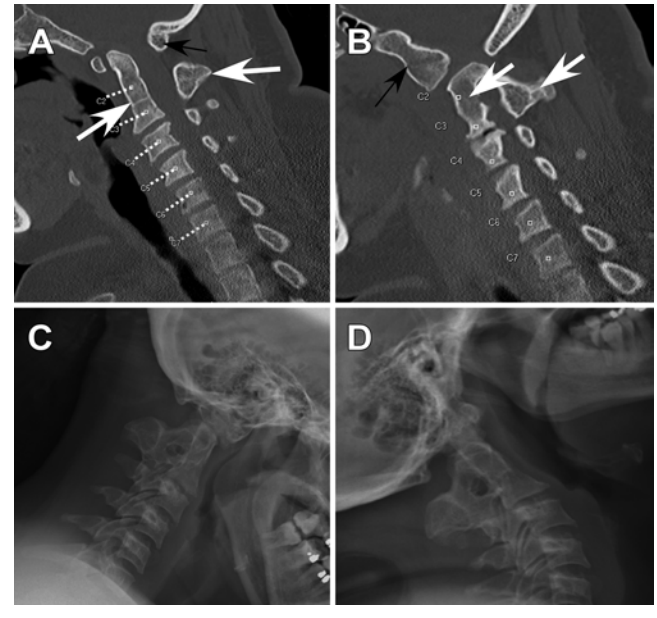


Fig. 2. Computed tomography scans and dynamic radiographs demonstrating an unstable congenitally fused occiput to C-1 and C-2 to C-3. **A:** Midline sagittal CT scan of the cervical spine. The *black arrow* highlights fusion of the occiput and C-1. The *white arrows* demonstrate fusion of both the anterior and posterior elements of C-2 and C-3. **B:** Parasagittal CT scan of the cervical spine. Once again, the *black arrow* highlights the fusion of the occiput and C-1. The *white arrows* again demonstrate anterior and posterior fusion of C-2 and C-3. **C:** Lateral radiograph of the cervical spine in flexion demonstrating anterior translation of the fused occiput-C1 relative to the C2–3 fusion mass. **D:** Lateral radiograph of the cervical spine in extension demonstrating posterior translation of the fused occiput-C1 relative to the C2–3 fusion mass.

Postoperative Course. The patient was successfully extubated on the day following the procedure and had a repeat angiogram on postoperative Day 3 that showed no evidence of restricted flow with flexion or extension of the neck. A cervical collar was maintained and the patient remained neurologically intact. Four weeks postopera-

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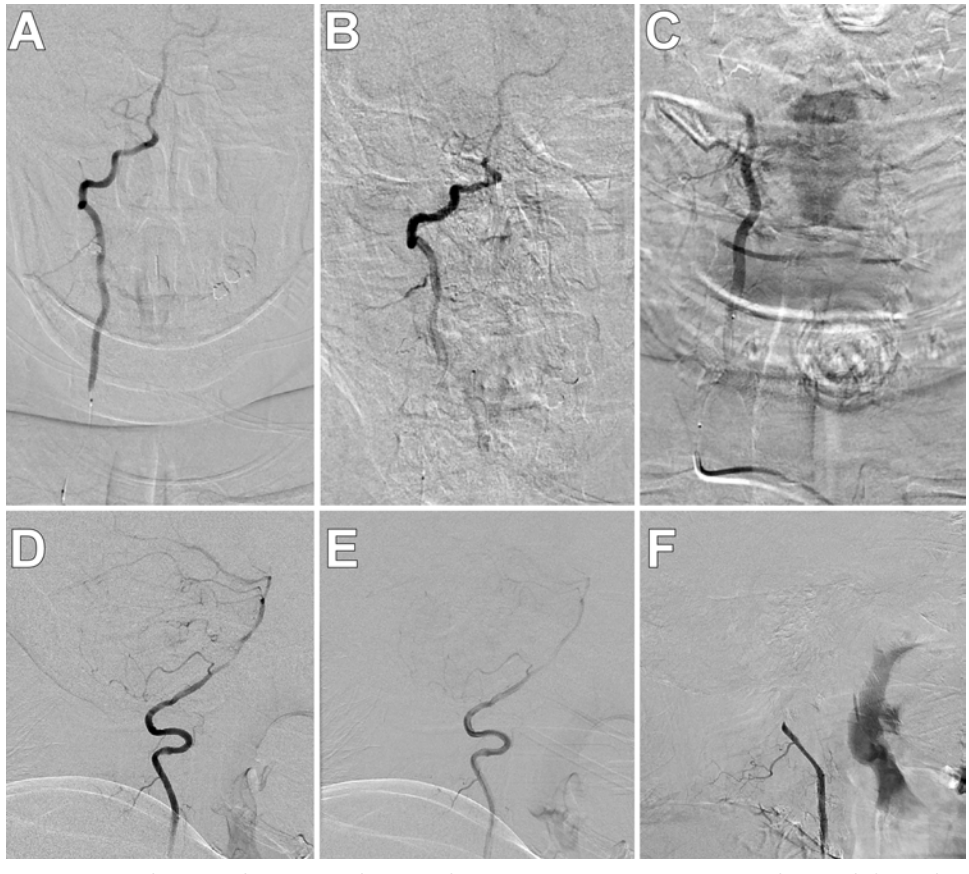


Fig. 3. Anteroposterior (upper row) and lateral (lower row) right VA angiograms in the neutral (A and D), flexed (B and E), and slightly extended (C and F) neck positions. **A and D:** Images obtained in a neutral neck position demonstrating brisk filling of the basilar system. **B and E:** Images obtained in a flexed neck position demonstrating filling of the basilar system. **C and F:** Images obtained in the slightly extended neck position demonstrating an abrupt cessation of flow in the V_3 segment of the VA.

tively she was doing well without swallowing difficulties or any new neurological complaints. She continues to do well without neurological or swallowing complaints at 15 months postoperatively.

Discussion

We report a unique case of a woman without a history of trauma who presented with symptoms of vertebrobasilar insufficiency (VBI) and was found to have a congenital cervical spine fusion with resultant dynamic vertebral occlusion demonstrated on cerebral angiography. Although previous cases of VA occlusion have been described with physiological head rotation, none have been previously associated with flexion or extension. This has been studied by Brown and Tatlow who performed postmortem VA angiograms on 41 cadavers and could not produce VA occlusion on severe extension or flexion.[4] Only with extension combined with rotation to 90° could they produce complete occlusion of the VA.

Vertebral artery stenosis or occlusion due to physiological rotation is a rare entity,[10] and the current presentation has not been reported in the literature to date. Given these relatively uncommon entities, patients and their physicians may not recognize that their symptoms are associated with certain physiological head movement. Therefore, it is imperative that patients presenting with symptoms of VBI, especially younger patients with no risk factors for stroke, be questioned on the association with head position. In patients presenting with VBI symptoms associated with head position, we recommend dynamic bone and vascular imaging be obtained.

In most previous cases of VBI due to VA stenosis, arterial occlusion tended to occur in the dominant VA and was secondary to mechanical compression, stretching, or subluxation of the cervical spine. In rotational occlusion, the most common area for compression is at the C1–2 junction. It is postulated that this occurs because of a fixed atlantoaxial joint on the side to which the head is rotated. The contralateral facet then stretches or sometimes subluxes, which causes a stretching of the VA in the foramen transversarium between C-1 and C-2.[2,22,27,28,33,36] It is well known that occlusion or stenosis of one VA is usually asymptomatic and rarely leads to clinical symptomology if the other VA or anterior circulation is able to compensate.[36] In our case, however, we postulate one of two hypotheses. First, we have demonstrated that minimal extension of the neck occluded 1 VA (Fig. 3) and in addition likely restricted the flow that remained in the contralateral VA. Perhaps the remaining flow was insufficient to maintain cerebral perfusion, producing her symptoms and stroke. A second plausible hypothesis is

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that with more pronounced neck extension (not tested on cerebral angiography due to concern of worsening her strokes), both VAs may have been occluded, causing her symptoms and strokes. These 2 hypotheses are supported by the fact that our patient suffered both ipsilateral and contralateral strokes at different times of her presentation and we did not formally test for dynamic compression of her left VA with further extension or addition of rotation to her neck.

Given that our case has a unique pathophysiological process, management strategies for treatment have not been well studied. However, several studies have investigated the management of rotational VA occlusion due to C1–2 rotational compression. Conservative management therapies have been attempted in rotational VA occlusion, including cervical collars to restrict head and neck rotation, the use of anticoagulant or antiplatelet therapies, the application of cervical traction, and behavioral modification.[12,22] Although there are no studies directly comparing these strategies to surgical treatment, most authors state that conservative therapy is usually not viable in these cases. Surgical treatment has been performed and generally includes either decompression of the VA at the site of compression, fusion of the spine in a noncompressed state, or the employment of these 2 strategies concomitantly.[1,6,13,14,24,31] A study by Matsuyama et al.[21] reported on treatment of 17 patients with bow hunter's stroke, 8 of whom had a posterior C1–2 fusion. The patients who underwent fusion appeared to have longer-term patency of their VA as well as remained asymptomatic neurologically when compared with patients who underwent decompression only. When making a decision on the appropriate surgical approach (anterior decompression and fusion vs posterior decompression and fusion), one should consider the site of compression and whether there is mainly an anterior compression (overgrowth of the uncinate process, large anterior osteophytes, or other anterior compression) or posterior compression due to facet hypertrophy. In addition, one should always keep in mind that posterior fusions at the most commonly affected level (C1-2) will likely cause loss of head rotation of greater than 50% and have significant other morbidities. Finally, one must consider whether there is an instability component to the compression; if so, fusion of the unstable segments as opposed to decompression alone is usually warranted.

Our patient presented with congenital anomalies of the craniocervical junction as well as the subaxial spine. On dynamic flexion and extension views (Fig. 2), instability was noted between the occiput-C1 fusion and C2–3 fusion masses. We postulate that with cervical extension, the VA was stretched and occluded. Given the instability and vascular compression at the craniocervical junction as well as normal flow in a neutral position, we chose to manage this using a stabilizing occiptocervical fusion in a slightly flexed position to not compromise the VA while allowing minimal morbidity to swallowing function in our patient. Immediate postoperative CT angiogram as well as a delayed cerebral angiogram were obtained and demonstrated appropriate flow within the vertebrobasilar system. The patient remains clinically well and has not had further symptoms. She has recovered well from the surgery without any swallowing morbidity. Long-term follow-up will be necessary to assess both the potential swallowing/breathing difficulties as well as increasing kyphosis that may result from fusion of a young patient in slight flexion.

In conclusion, we present the first case of VA occlusion caused by physiological extension of the neck as well as a relevant review of the literature on this rare topic. Further case reports and series are warranted on this topic to elucidate the appropriate diagnostic and treatment strategies needed for these patients.

Disclosure

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Author contributions to the study and manuscript preparation include the following. Conception and design: Hwang, Safain, Malek. Acquisition of data: Hwang, Safain, Malek. Analysis and interpretation of data: all authors. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Hwang. Study supervision: Hwang, Safain, Malek.

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