Case Report

Occipital Bone Abnormality Causing Recurrent Posterior Circulation Strokes

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Abstract—We report the case of a young man with recurrent posterior circulation strokes over the course of 6 years. Standard stroke evaluation was unremarkable until careful review of catheter angiogram and CT angiogram images revealed a bony protuberance from the occiput impinging on the left vertebral artery. Local vessel injury with thrombosis and distal embolization is the presumed etiology of the recurrent infarcts. Surgical removal of this developmental anomaly was accomplished, with no subsequent neurological events. (Stroke. 2011;42:e370-e372.)

Key Words: anatomy ■ angiography ■ neurosurgery ■ stroke ■ vertebrobasilar

A 26-year-old man with recurrent strokes came to our neurology clinic for a second opinion. The patient was an otherwise healthy man whose first neurological event was at age 20 when he presented to an outside hospital with vertigo and left hemiparesis, which had been preceded by a severe headache for ≈2 weeks. Imaging revealed infarcts in the left thalamus, left occipital lobe, and bilateral cerebellum (the largest area in the territory of the left posterior inferior cerebellar artery; Figure 1A). Angiogram revealed a distal left posterior cerebral artery cut-off, but no other irregularities. Further evaluation was unremarkable, including evaluation for autoimmune or hypercoagulable state and lipid panel. Transesophageal echocardiogram showed a small patent foramen ovale by color Doppler, but without right-to-left shunt of saline bubbles. He was discharged with warfarin.

The next week, dysarthria, vertigo, nausea, vomiting, and ataxia developed. His international normalized ratio was therapeutic at 2.2. MRI revealed small, new, acute infarcts in bilateral cerebellar hemispheres (Figure 1Bi). Angiogram showed distal occlusions in branches of the left posterior inferior cerebellar artery, left posterior cerebral artery, and right superior cerebellar artery. CT angiogram noted a bony anomaly of C1 on the right forming a narrow channel with focal narrowing of the right vertebral artery. The large stroke in the left posterior inferior cerebellar artery territory made it unlikely that this narrowing in the right vertebral artery was causative. He was again discharged with warfarin without a clear etiology of the infarcts.

He recovered well, and 2 years later (age 22) he was switched from warfarin to aspirin; the next year, aspirin was discontinued. At age 24, he experienced a transient episode of left homonymous hemianopsia, and 2 weeks later diplopia, vertigo, ataxia, and headache occurred. MRI showed acute right occipital infarcts (Figure 1Bii). He was again prescribed warfarin.

He was asymptomatic until 2.5 years later (age 26), when gait instability developed the day after riding a roller coaster. MRI showed no new lesions. Five days later, diplopia and left hemiparesis developed. MRI revealed new strokes in the right anterior cerebellar hemisphere, right thalamus, and left posterior temporal lobe (Figure 1Biii). Angiogram was thought to be unremarkable. Two weeks later, numbness and weakness of his left hemibody with new bilateral thalamic infarcts developed (Figure 1Biv).

At that point, he presented to our clinic for another opinion on the etiology of these recurrent events. Doppler ultrasound with head-turning maneuvers did not reveal any significant velocity changes. Further review of the recent angiogram identified left vertebral artery dilation at C1 (Figure 2A). This was further evaluated with CT angiogram with 3D reconstructed images. Again seen was the diffusely small, right vertebral artery coursing through a tight osseous channel formed by a bifid posterior C1 arch. Additionally, adjacent to the fusiform widening of the left vertebral artery at C1, a bony protuberance extending from the occiput was identified (Figure 2B, C). With retrospective review, this bone protrusion was present on previous imaging, but the dangerous position relative to the artery was more apparent on coronal sections and 3D reconstructions. We concluded that the etiology of his strokes was recurrent intimal injury in the left vertebral artery attributable to compression by this adjacent bone causing thrombi formation with distal embolization.

The findings were discussed with the patient and it was decided to surgically remove the anomalous bone. A “hockey stick” incision was made from C2 to the torcula and lateral to the ear. The suboccipital muscles were mobilized and retracted laterally; using boney landmarks and Stealth CT navigation, the suboccipital area, arch of C1, and vertebral...
artery were identified. We decided on this extensive exposure instead of a small muscle splitting procedure to have a wider view to minimize potential damage to the vertebral artery. Using the operative microscope, the artery was exposed and protected, and the anomalous bone was identified superiorly originating from the occipital plate. Even in the flexed position, this boney protrusion was pressing against the vertebral artery. Intraoperative Doppler ultrasound was used throughout the case to verify location of the artery. Bone was removed with a high-speed drill until flush with the occipital plate.

Figure 1. Recurrent posterior circulation infarcts. A, Fluid-attenuated inversion recovery sequence from second presentation at age 20, showing all previous areas of infarction. B, Diffusion-weighted imaging from (i) second presentation at age 20; (ii) age 24; (iii) age 26; and (iv) age 26, 2 weeks later. All lesions are in the left vertebral artery distribution.

Figure 2. Imaging of vertebral arteries, C1, and occiput. A, Angiogram of left vertebral artery injection. Left, age 20; right, age 26. Arrow indicates area of vessel damage with fusiform widening seen on the later image. B, Preoperative coronal CT angiogram. Arrowhead indicates right vertebral artery course through C1 anomalous bony channel. Arrow indicates left vertebral artery with adjacent boney protrusion from the occiput. C, CT angiogram 3D reconstructions of the left vertebral artery course over C1. Removal of the occiput protruberance and vessel caliber normalization on 3-month postoperative image (right).
plate. After removal, the vertebral artery assumed a more superior course, pulsating well and free of any compression.

The patient recovered well with no postoperative events and was discharged with warfarin. CT angiogram 3 months later showed normalization of artery caliber (Figure 2C). Some slight past pointing on left finger-to-nose testing was the only deficit remaining on examination. He denied any new episodes and also noted that he had previously had a chronic low-grade headache and muscle tightness in his neck, which had resolved. He was switched to dual antiplatelets (aspirin 81 mg and clopidogrel 75 mg daily) for 3 months and then continued aspirin alone. He remains symptom-free 2 years later.

**Discussion**

Typically, the vertebral artery exits the C1 transverse foramen and runs in a sulcus over the C1 posterior arch before entering the foramen magnum. This sulcus is sometimes fully or partially bridged superiorly or laterally by anomalous bone development of C1. These bony structures have been given many names, including foramen arcuale and posterior lateral ponticuli.1,2 In many vertebrates, the vertebral artery runs in a bony tunnel in this area, and it is speculated that this structure provided attachment surfaces for extensor head muscles and subsequently disappeared with the transition to vertical loading of head weight with upright posture. Human case series have found complete or partial presence of this tunnel in 1% to 37%.2 At times, presence of this tunnel is associated with vertebral artery narrowing and symptoms of posterior circulation hypoperfusion. Our patient has such a structure on the right side, with narrowing of the vertebral artery. However, the left posterior inferior cerebellar artery infarct suggests against a right vertebral etiology. The left abnormality is in a similar anatomic location superior to the vertebral artery but arises from the occipital bone rather than C1. It is presumed that this structure also represents an anomalous developmental remnant from formation of the C1 arch. To our knowledge, such a structure protruding from the occipital bone causing vertebral artery pathology has not been previously described.

This case demonstrates that with infarcts of unclear etiology, localization to determine the vascular distribution of lesions and careful review of imaging may reveal uncommon anatomic variants as possible causes. CT angiogram is a useful tool that gives detailed views of vessel caliber and surrounding structures. One can review the anatomy along the length of vessels in all planes of the section and with 3D reconstructions, allowing for a fuller understanding of any structural pathology. This is especially important because one may find lesions that are amenable to intervention to prevent further infarcts, as was the case with our patient.

**Disclosures**

None.

**References**

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Stroke. 2011;42:e370-e372; originally published online March 24, 2011;
doi: 10.1161/STROKEAHA.110.612176
Stroke is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
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Print ISSN: 0039-2499. Online ISSN: 1524-4628

The online version of this article, along with updated information and services, is located on the
World Wide Web at:
http://stroke.ahajournals.org/content/42/5/e370

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