Bow hunter's syndrome due to an embolic mechanism: illustrative case

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BACKGROUND Bow hunter's syndrome (BHS) is an uncommon cause of vertebrobasilar ischemic stroke, which results from occlusion or injury to the vertebral artery (VA) during neck rotation. Although hemodynamic insufficiency is the predominant underlying mechanism of this entity, BHS due to embolic mechanisms is rare. The authors report a case of BHS characterized by repeated posterior circulation embolism and present some considerations of BHS with an embolic mechanism.

OBSERVATIONS A 57-year-old man suffered from repeated embolic stroke in the posterior circulation. Digital subtraction angiography revealed caliber irregularity of the V3 segment of the left nondominant-side VA, which occluded when the neck rotated to the right side. The patient was diagnosed with BHS with an embolic mechanism due to endothelial damage caused by osteophytes at the C1 foramen transversarium. After C1–C2 fusion surgery, the patient never experienced the recurrence of stroke. According to a literature review, BHS due to embolic mechanisms tends to occur in young male adults, manifesting as recurrent stroke in the posterior circulation. Involvement of the nondominant-side VA can cause BHS with an underlying embolic mechanism.

LESSONS BHS due to an embolic mechanism should be considered as a differential diagnosis if patients have repeated embolic strokes in the posterior circulation.

<https://thejns.org/doi/abs/10.3171/CASE20150>

KEYWORDS bow hunter's syndrome; embolic stroke; posterior circulation; vertebrobasilar insufficiency

Bow hunter's syndrome (BHS) is an uncommon cause of vertebrobasilar ischemic stroke. In 1978, Sorensen coined the term "bow hunter's syndrome" to describe a case of vertebrobasilar ischemic stroke resulting from the occlusion of or injury to the vertebral artery (VA) during neck rotation.^{1–3} Although most cases with BHS manifest as transient ischemic attacks or watershed infarctions due to hemodynamic insufficiency, BHS due to embolic mechanisms is rare. It has been postulated that repeated neck rotation and VA compression against the surrounding structure may cause injury to the vessel wall, which leads to direct injury of the intimal lining, arterial dissection, or pseudoaneurysm formation, resulting in subsequent thrombus formation and distal embolism.^{3,4}

To date, no reports have discussed BHS due to embolic mechanisms in detail. We herein report a case of BHS characterized by repeated posterior circulation embolism and present some considerations of BHS due to embolic mechanism with a literature review.

Illustrative Case

A 57-year-old man with a past history of hypertension and dyslipidemia suffered from repeated cerebral infarction in the posterior circulation territory 4 times in the past 4 years (Fig. 1). He complained of dizziness when he turned his neck to the right, but he never experienced syncope or visual disturbance. He was medicated with aspirin after the onset of the first infarction, and together with warfarin after the second attack, although the underlying mechanism behind the repeated cerebral infarction had not been determined. After the onset of the fourth attack, his primary care physician ordered magnetic resonance angiography (MRA), which showed no abnormalities in the neutral head position. However, MRA performed during neck rotation to the right revealed stenosis of the left VA, which led to the suspicion of BHS. He was referred to our hospital for further examination and treatment. Digital subtraction angiography showed stenotic change as well as caliber irregularity at the V3 segment of the left VA in the neutral

ABBREVIATIONS BHS = bow hunter's syndrome; CT = computed tomography; MRA = magnetic resonance angiography; VA = vertebral artery.

INCLUDE WHEN CITING Published February 15, 2021; DOI: 10.3171/CASE20150.

SUBMITTED December 10, 2020. **ACCEPTED** December 17, 2020.

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FIG. 1. Diffusion-weighted images at the first attack 4 years ago (**A**), at the second attack 3 years ago (**B**), at the third attack 3 months ago (**C**), and at the fourth attack at the time of referral to our department (**D**). The patient's ischemic stroke recurred only in the posterior circulation.

head position (Fig. 2A). While rotating the neck to the right, the contrast agent stagnated at the left stenotic VA of the C1–C2 level (Fig. 2B). Cone-beam computed tomography (CT) revealed an osteophyte at the entry of the foramen transversarium on the left side of C1, which was assumed to compress the left VA (Fig. 2C and D). Three-dimensional CT angiography demonstrated that the right (contralateral) VA had developed similar to the left one. Based on these findings, he was diagnosed with BHS due to an embolic mechanism.

He underwent fusion surgery for C1–C2 (Magerl's method). Fixing the C1–C2 joint, mechanical compression on the left VA was reduced (Fig. 3). Postoperatively, the patient had no new neurological deficits, and he was discharged with no complications. No recurrence of stroke or dizziness was observed for more than 1 year postoperatively.

Results of the Literature Review

To elucidate the characteristics of BHS due to embolic mechanisms, we performed a literature review as follows. After we searched the publications on PubMed with the keywords “bow hunter syndrome” and “bow hunter's syndrome,” the cases with BHS due to embolic mechanism were picked up by the image findings of ischemic embolic stroke or the description in the text.

There were 22 patients with BHS due to embolic mechanisms, including the present case, as shown in Table 1.^{3,5–18} The overall mean age of the patients was 31 ± 22 years (Table 2). BHS due to an embolic mechanism was observed in 20 men and 2 women. The dominant and nondominant sides of the VA were affected equally. Recurrence of ischemic stroke was observed in 19 cases, and the number of patients whose embolic source was determined by angiography was 13. The most common site of VA compression was C1, followed by C2, the C1–C2 joint, and the craniocervical junction. Bone spur and osteophytes were the most common underlying etiologies, which were diagnosed with image findings. Surgery was usually chosen to counteract BHS with an embolic mechanism (18 cases) in contrast to conservative therapy performed in only 3 cases. Spinal fusion surgery was most commonly performed, followed by decompression surgery and endovascular surgery.

FIG. 2. Preoperative dynamic digital subtraction angiography of the left VA in the neutral head position (**A**) and in the rotated neck position (**B**). Note the stenosis and caliber irregularity of the V3 segment (white arrow) and stagnation of the contrast agent while rotating the neck to the right (white arrowhead). Axial (**C**) and sagittal (**D**) cone-beam CT images reveal that the left VA is compressed by an osteophyte of C1 (white arrows).

Discussion

Observations

BHS was originally a descriptive synonym for rotational verte-brobasilar insufficiency caused by hemodynamic mechanisms.¹⁹ Previous reports revealed that the mean age of onset for BHS was 53–57 years, and the male-to-female ratio was about 2:1.^{2,20} The prevalent symptoms are syncopal or near-syncopal events, drop attacks, vertigo, dizziness, and impaired vision upon neck rotation.^{2,21} A common characteristic of conventional BHS is that these symptoms are usually transient and reversible.^{2,20} In contrast to BHS due to hemodynamic insufficiency, the summarized data in the present study enables us to elucidate the different characteristics of BHS due to embolic mechanisms. According to the literature review, the mean age of onset was 31 ± 22 years, and the male-to-female ratio was 6:1. Thus, younger male adults are more likely to have BHS with embolic mechanisms than BHS due to hemodynamic insufficiency. In addition, almost all patients with BHS due to an embolic mechanism suffered from recurrence of ischemic stroke in the posterior circulation territory. Most cases have permanent neurological deficits and are diagnosed at

FIG. 3. Postoperative lateral images of cervical radiography (**left**) and angiography (**right**). Note that C1–C2 fixation improves stenosis of the left VA.

the time of recurrent strokes. Although the dominant-side VA is commonly affected in BHS with hemodynamic insufficiency,^{2,3} involvement of the nondominant-side VA can also be a cause of BHS due to an embolic mechanism. These characteristic discrepancies between conventional BHS and BHS due to an embolic mechanism might be explained by their difference in the underlying pathology. In BHS due to hemodynamic insufficiency, because rotation of the neck causes dynamic and reversible occlusion of the VA in addition to the lack of collateral blood supply to the brainstem, the symptoms are usually transient. Conversely, BHS with an embolic mechanism may be caused by endothelial damage due to vascular compression by the surrounding bony structures during neck rotation, leading to thrombus formation and subsequent embolism. Therefore, even the involvement of the nondominant-side VA can result in repeated ischemic stroke in this type of BHS. Based on these findings, in cases in which patients have repeated embolic strokes in the posterior circulation, detailed examination should be conducted to identify BHS due to embolic mechanisms.

Dynamic angiography is the gold standard for the diagnosis of BHS because normal neutral vascular imaging does not preclude the diagnosis of BHS. The patent arteries in the neutral neck position, as well as stenotic or occlusive arteries in the rotated position, strongly suggest a diagnosis of BHS. Furthermore, CT, magnetic resonance imaging,

TABLE 1. Cases of BHS due to an embolic mechanism

Author, Year	Age (yrs)/Sex	Location	VA Dominance	Emboli Source	Underlying Pathology	Recurrent Stroke	Treatment
Lu DC et al., 2009 ⁵	12/M	Occipital–C1	Dominant	Dissection	Bone spur	+	Decompression
Greiner HM et al., 2010 ⁶	15/M	C1	Dominant	Dissection	Congenital bony anomaly	+	Decompression
Anene-Maidoh TI et al., 2013 ⁷	16/M	C1	Dominant	Dissection	Congenital bony anomaly	+	Decompression
Safain MG et al., 2014 ⁸	73/M	C1–C2	Nondominant	N/A	Congenital bony anomaly	+	Fusion
Thomas B et al., 2015 ⁹	60/M	C5	Nondominant	Wall injury	Osteophyte	+	PAO
Yamaguchi S et al., 2014 ¹⁰	45/M	C1–C2	Dominant	Wall injury	VA fenestration	+	Fusion
Kageyama H et al., 2016 ¹¹	17/M	C1	Dominant	Dissection	Severe traction by repeated hyper-rotation	+	Fusion
	18/M	C2	Dominant	Dissection	Severe traction by repeated hyper-rotation	+	Fusion
Yagi K et al., 2017 ¹²	74/M	C4–C5	Dominant	N/A	Osteophyte	+	Decompression
Berti AF et al., 2018 ¹³	56/M	C5	Nondominant	Dissection	Idiopathic	+	PAO
Fujii M et al., 2020 ¹⁸	16/M	Occipital–C1	N/A	Dissection	Bone spur	+	N/A
Jadeja N & Nalleballe K, 2018 ³	24/M	C2	Dominant	Pseudoaneurysm	Congenital bony anomaly	+	Medication
Kameda T et al., 2018 ¹⁴	56/M	C1	Dominant	N/A	Osteophyte	–	Decompression
Saadi A & Klein JP, 2018 ¹⁵	19/M	C2	Dominant	Dissection	Neck muscle hypertrophy	+	Decompression
Takeshima Y et al., 2018 ¹⁶	34/M	C1	Nondominant	N/A	Congenital bony anomaly	–	Fusion
	7/M	C2	Nondominant	N/A	Atlantoaxial subluxation	+	Fusion
	22/M	C1	Dominant	N/A	Idiopathic	+	Fusion
	52/M	C2	Dominant	N/A	Atlantoaxial subluxation	+	Fusion
	16/M	C1	Nondominant	N/A	Idiopathic	–	Medication
	18/F	C2	Nondominant	N/A	Idiopathic	+	Fusion
Cohen NT et al., 2020 ¹⁷	2/F	C1–C2	Nondominant	Dissection	Atlantooccipital ligament calcification	+	Medication
Present case	57/M	C1–C2	Nondominant	Wall injury	Osteophyte	+	Fusion

+ = yes; – = no; N/A = not available; PAO = parent artery occlusion.

TABLE 2. Summary of cases with BHS due to an embolic mechanism

Characteristic	Value
Total no. of patients	22
Age, yrs (mean $\pm$ SD)	31.0 $\pm$ 22.2
Sex, M/F, no. (%)	20 (91)/2 (9)
Dominant-side VA affected, no. (%)	11 (50)
Recurrent stroke, no. (%)	19 (86)
Angiographic diagnosis of source of embolism, no. (%)	13 (59)
Compression level of VA, no. (%)	
Occipital–C1	3 (14)
C1	6 (27)
C1–C2	4 (18)
C2	6 (27)
C4–C5	1 (5)
C5	2 (9)
Underlying pathology, no. (%)	
Idiopathic	4 (18)
Bone spur/osteophyte	7 (31)
Congenital bony anomaly	4 (18)
Atlantoaxial subluxation	2 (9)
Severe traction by repeated hyper-rotation	2 (9)
VA fenestration	1 (5)
Neck muscle hypertrophy	1 (5)
Atlantooccipital ligament calcification	1 (5)
Treatment modality, no. (%)	
Fusion surgery	10 (48)
Decompression surgery	5 (24)
Endovascular surgery	3 (14)
Conservative treatment (medication)	3 (14)

SD = standard deviation.

and neurosonography are reported to be important for the detection of cerebral infarction or stenotic arteries.²¹ These examinations are also useful in detecting the underlying etiology of VA compression, such as osteophytes, bone spurs, atlantoaxial instability, cervical disc herniation, and congenital bony anomalies.^{2,3,5,6,9,10,15,17} In contrast to BHS due to hemodynamic insufficiency, vessel wall injury, such as stenosis, dissection, and pseudoaneurysm at the compression site, is the key finding for the diagnosis of BHS due to an embolic mechanism, which can usually be detected with angiography. In the present case, dynamic digital subtraction angiography revealed stenotic VA changes in the neutral head position as well as VA occlusion in the rotated neck position. In addition, cone-beam CT revealed osteophytes at the entry of the foramen transversarium, which presumably induced vessel wall injury in the VA by mechanical compression. These radiological findings led us to the diagnosis of BHS due to an embolic mechanism. Therefore, we strongly recommend performing dynamic angiography to detect any evidence of vessel wall injury, as well as cone-beam CT to examine the surrounding structures of the affected VA, in case BHS due to an embolic mechanism is suspected.

Treatment of BHS varies widely depending on the location and pathology of the occlusion.² Although conservative therapy is chosen in a small number of cases,¹⁹ surgical therapy carries an excellent

prognosis and includes cervical spine fusion, cervical decompression, and endovascular treatment such as stenting and parent artery occlusion. According to our literature review, 18 out of 21 cases of BHS due to an embolic mechanism were surgically treated. Cervical spine fusion, which is most commonly performed, is considered to be highly curative because of the complete prevention of postoperative vascular compression and subsequent vessel wall injury.¹⁶ Patients with recurring symptoms would be good candidates, although the postoperative limitation of neck rotation is the disadvantage of cervical fusion surgery. Decompression surgery is another option that can be performed in patients with obvious preoperative vascular compression by anatomical structures. Despite the potential risk of intraoperative VA injury, as well as the risk of restenosis, decompression surgery would not limit head rotation postoperatively.²¹ Endovascular treatment is a less invasive option for BHS treatment. Stenting and parent artery occlusion are reported to be effective in patients with BHS with embolic mechanisms.^{22,23} If the affected VA is on the nondominant side, parent artery occlusion should be considered as a radical treatment. In our case, we chose cervical fusion surgery because the patient had repeated ischemic strokes, necessitating a curative treatment. Appropriate BHS management should be determined on a case-by-case basis considering the location and pathology of the disease.

Lessons

BHS due to an embolic mechanism is a rare entity that mainly develops in young male adults, manifesting as recurrent strokes in the posterior circulation. In addition, involvement of the nondominant-side VA can cause this rare entity. Given our experience and the literature review, we propose that BHS due to an embolic mechanism should be considered as a differential diagnosis if patients have repeated embolic strokes in the posterior circulation territory.

Acknowledgments

This work was supported by MEXT | Japanese Society for the Promotion of Science (grant no. JP19K18414; Dr. Sakata).

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Sakata, Shingai, Suzuki, Ezura, Tominaga. Acquisition of data: Sakata, Shingai, Endo, Suzuki. Analysis and interpretation of data: Sakata, Shingai, Endo, Suzuki. Drafting the article: Sakata, Shingai, Suzuki. Critically revising the article: Sakata, Endo, Suzuki. Reviewed submitted version of manuscript: Sakata, Endo, Suzuki, Tominaga. Approved the final version of the manuscript on behalf of all authors: Sakata. Statistical analysis: Suzuki. Administrative/technical/material support: Suzuki, Ezura. Study supervision: Suzuki, Tominaga.

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