Korean J Neurotrauma. 2022 Oct;18(2):410-417
https://doi.org/10.13004/kjnt.2022.18.e63
pISSN 2234-8999·eISSN 2288-2243

KJNT

Case Report

Check for updates

Rotational Vertebral Artery Syndrome (Bow Hunter's Syndrome): A Rare Differential Diagnosis in Patients With Syncope

In-Seo Hong [1], Eun-Ho Jung [1], Kyung Hwan Kim [1], Hyon-Jo Kwon [1], Seung-Won Choi [1], Seon-Hwan Kim [1], Hyeon-Song Koh [1], Jin-Young Youm [1], and Han-Joo Lee [1,2]

[1]Department of Neurosurgery, Chungnam National University Hospital, Chungnam National University School of Medicine, Daejeon, Korea
[2]Department of Neurosurgery, Yonsei University College of Medicine, Seoul, Korea

OPEN ACCESS

Received: Sep 12, 2022
Revised: Oct 9, 2022
Accepted: Oct 10, 2022
Published online: Oct 24, 2022

Address for correspondence:
Han-Joo Lee
Department of Neurosurgery, Chungnam National University Hospital, Chungnam National University School of Medicine, 282 Munhwa-ro, Jung-gu, Daejeon 35015, Korea.
Email: metalblue0205@hotmail.com

ORCID iDs
In-Seo Hong
https://orcid.org/0000-0002-4773-8921
Eun-Ho Jung
https://orcid.org/0000-0003-4326-8499
Kyung Hwan Kim
https://orcid.org/0000-0003-1244-6969
Hyon-Jo Kwon
https://orcid.org/0000-0003-1077-2461
Seung-Won Choi
https://orcid.org/0000-0001-8610-887X
Seon-Hwan Kim
https://orcid.org/0000-0002-5600-2801

ABSTRACT

Syncope is a common symptom in clinical practice. Rotational vertebral artery occlusion syndrome, also referred to as Bow Hunter's syndrome (BHS), is a rare condition associated with syncope and is caused by mechanical occlusion or stenosis secondary to mechanical compression of the vertebral artery during head rotation. BHS is associated with a multifactorial etiology; however, in most cases, this condition is attributed to degenerative changes. A 53-year-old man visited our hospital for the evaluation of fainting and dizziness episodes that occurred when he turned his head. Evaluation as an outpatient in the Department of Neurology showed a positive result on the Frenzel goggle test. Transfemoral cerebral angiography performed at the Department of Neurosurgery revealed stenosis of the proximal right vertebral artery. Complete occlusion of the vertebral artery was observed, and the head was turned to the right. Decompression and fusion were performed, and the contributory lesion was completely removed. Postoperative imaging confirmed complete removal of the spur and sufficient vertebral artery decompression; the patient's symptoms resolved postoperatively.

Keywords: Vertebral artery insufficiency; Syncope; Vertebral artery stenosis

INTRODUCTION

Syncope, with a lifetime prevalence of 20%–40% is commonly encountered in clinical practice.[5] Etiologically, syncope can be categorized into cardiovascular and non-cardiovascular syncope. Non-cardiovascular syncope includes that associated with neurological conditions (seizures, cerebrovascular accidents, and autonomic neuropathy), hypoglycemia, and miscellaneous conditions (pulmonary embolism, psychogenic disorders).[6,14]

The blood circulation system of the brain is divided into an anterior circulatory system that leads to the anterior cerebral artery and a middle cerebral artery through the carotid artery, and a posterior circulatory system that connects from 2 vertebral arteries to form a basilar artery.

Hyeon-Song Koh
https://orcid.org/0000-0003-2659-5535
Jin-Young Youm
https://orcid.org/0000-0001-9609-0415
Han-Joo Lee
https://orcid.org/0000-0003-4518-9057

Conflict of Interest
The authors have no financial conflicts of interest.

Among them, the term vertebrobasilar insufficiency (VBI) was used collectively for various causes that can cause ischemia in hemodynamics in the posterior circulation. The term VBI includes various diseases that can cause posterior circulatory ischemia, such as subclavian steal syndrome and rotational vertebral artery syndrome as well as vertebrobasilar transient ischemic attack.

Rotational vertebral artery occlusion syndrome, also referred to as Bow Hunter's syndrome (BHS) is an extremely rare cause of neurological syncope and is attributed to mechanical occlusion or stenosis secondary to mechanical compression of the vertebral artery during head rotation. Symptoms of BHS include dizziness, nausea, dysarthria, headaches, syncope, vision disturbance, hearing loss, or drop attacks.[2,10,20] Occlusion is most commonly observed at the C2 level, and occlusion below this level is rare.[18,19]

BHS shows multifactorial etiology; however, degenerative changes (osteophytes, spondylosis, disc protrusion) are implicated as the most common cause (reported in 54% of all cases of BHS).[14] Osteophytes are predominantly observed at uncovertebral joints and originate from the anteromedial portion and compress the vertebral artery in a posterolateral direction in approximately 50% of patients. Osteophytes trigger inflammation that produces a fibrous band around the vertebral artery, which restricts the movement of the artery and causes obstruction during head rotation. In addition to rotation, neck extension causes obstruction.

Owing to the rarity of this condition, guidelines for diagnosis and treatment of BHS are unavailable, and BHS may be missed or misdiagnosed as a cause of syncope. We discuss the diagnostic and therapeutic approach to BHS in a patient with this rare condition that may precipitate syncope.

CASE REPORT

A 53-year-old man with a history of treated diabetes and hypertension and anterior cervical discectomy and fusion (C4/5) 2 years prior to presentation, visited our hospital for evaluation of a 2-month history of sudden onset of dizziness and syncope. The patient experienced loss of consciousness several times within 2 months and observed this symptom each time he turned his head to the right. The patient complained of a severe headache when he turned his head to the right during daily life, and although he could not remember the exact situation, he said that he immediately lost consciousness while turning his head to the right and maintaining the posture. The patient experienced immediate dizziness and loss of consciousness when he turned his head by >45° to the right. He also experienced occasional episodes of nausea and cold sweats. The patient was frequently exposed to noise owing to aircraft-related work and had partial hearing loss; however, he denied previous symptoms of dizziness or syncope.

The patient initially visited the otolaryngology outpatient department, and no ear-related abnormalities were detected; therefore, he was referred to a neurologist for treatment. The Frenzel goggle test performed as an outpatient showed positive findings when the patient turned his head to the right.

The patient was initially admitted to the neurology department for evaluation of syncope and experienced severe dizziness upon performing the Frenzel goggle test with his head to the right. Transfemoral cerebral angiography (TFCA) was recommended by the neurologist, and he underwent this imaging study upon transfer to the neurosurgery department.

https://doi.org/10.13004/kjnt.2022.18.e63

TFCA revealed stenosis of the proximal right vertebral artery without any left vertebral artery abnormalities (**FIGURE 1**). The right vertebral artery showed 48% stenosis with the patient placed in the supine position. We turned the patient's head to the right and performed the examination to confirm that head rotation led to his symptoms. We observed that head rotation to the right led to complete occlusion of the vertebral artery (**FIGURE 2**).

We performed cervical spine imaging for clearer delineation of the lesion. Radiography revealed C4/5 interbody fusion and a bone spur at the right C6/7 level; therefore, we performed computed tomography (CT) angiography, which revealed a large spur at the right C6/7 uncovertebral joint that led to vertebral artery compression (**FIGURE 3**). The spur was observed to project upward into the transverse foramen on axial CT views. Magnetic resonance imaging revealed no spinal cord compression; however, we observed foraminal stenosis at the right C6/7 level.

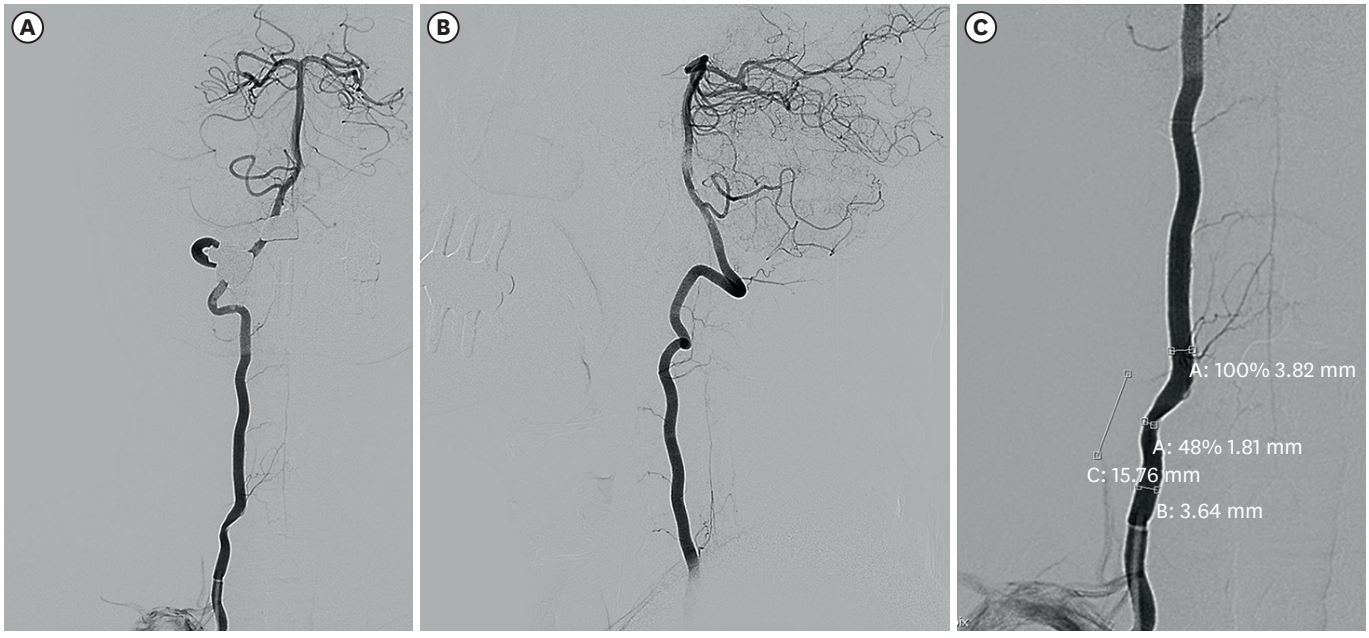

FIGURE 1. Transfemoral cerebral angiography on the right vertebral artery. (A) Anterior-posterior view. (B) Lateral view. (C) In anterior-posterior view, the right vertebral artery showed 48% stenosis.

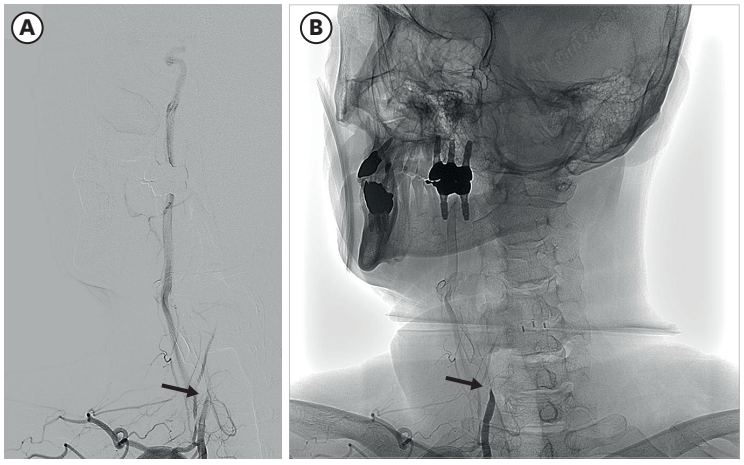

FIGURE 2. Transfemoral cerebral angiography on the right vertebral artery after turning the patient's head to the right. (A) General cerebral angiography. (B) Bone setting image. The arrow marks the stenosis point of the vertebral artery.

https://doi.org/10.13004/kjnt.2022.18.e63

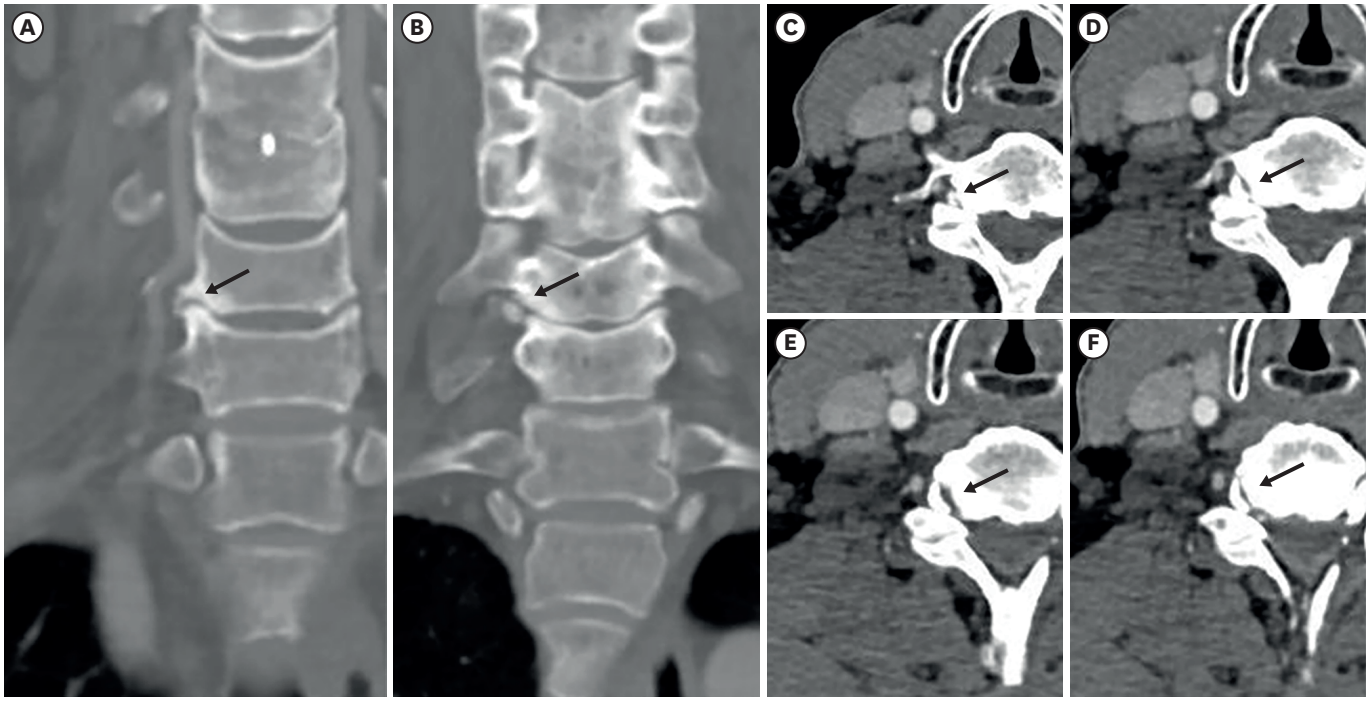

FIGURE 3. Computed tomography image with angiography for cervical spine. (A, B) Coronal image. (C-F) Axial image. The arrow indicates the point that the vertebral artery is pressed by osteophyte.

The patient opted to undergo surgery; we considered simple decompression or anterior fusion, and decided to proceed with anterior cervical fusion considering the case that provide insufficient decompression owing to high grown spur. We concluded that if sufficient fusion was successful, movement of the stenosis site would disappear and progression of stenosis could be prevented with head turning movements. We performed the usual anterior cervical discectomy and fusion procedure, with removal of the right uncinate process via a left-sided approach. A spur was observed posterior to the uncinate process, and the area surrounding the spur was carefully peeled off using a blunt hook (**FIGURE 4**). After confirming mobility of the spur, it was removed completely using a curette. Pulsation of the vertebral artery

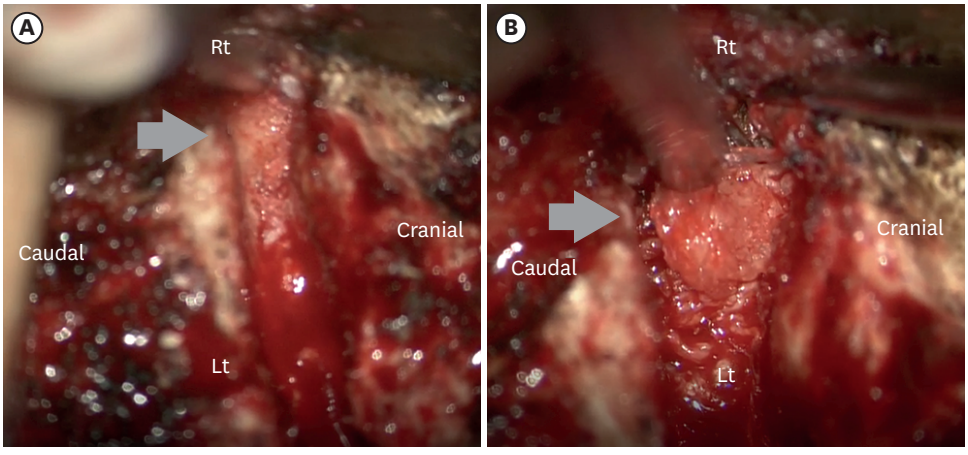

FIGURE 4. Intra-operative photography after removal of right uncinate process. (A) Gray arrow indicated osteophyte that compressed vertebral artery. (B) Gray arrows indicate the osteophytes lifted by blunt hooks. Lt: left, Rt: right.

https://doi.org/10.13004/kjnt.2022.18.e63

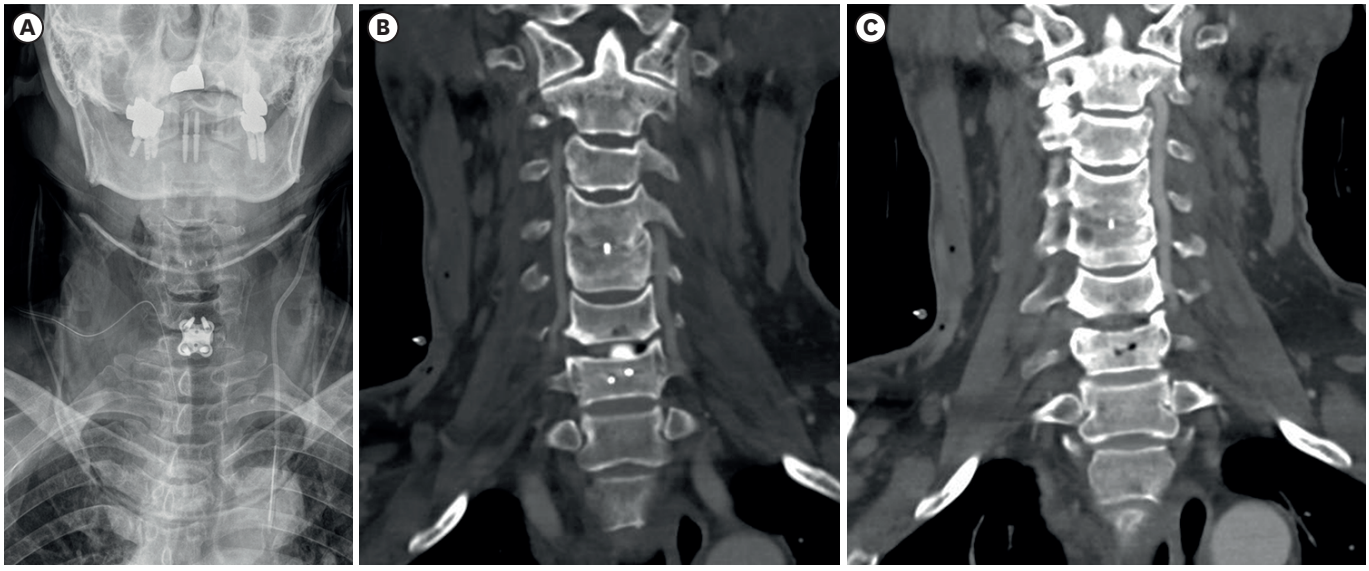

FIGURE 5. Post-operative image. (A) X-ray image immediately after surgery. (B, C) Coronal view of computed tomography image with angiography.

was confirmed, and we verified that the spur was appropriately removed from within the transverse foramen. We performed successful anterior fusion in this case.

Postoperative imaging confirmed complete removal of the spur with sufficient decompression of the vertebral artery (**FIGURE 5**). The patient's symptoms disappeared postoperatively and did not recur with head turning movements during follow-up a month later. The patient was followed up as an outpatient for 6 months and showed no abnormalities.

We summarized BHS-related papers published after year 2000, mainly referring to 6 reviews or case reports with literature review, and 94 original case reports.[1,7,8,12,15,17] One hundred eighteen patients were classified into 9 treatment methods, and they were classified into 3 groups according to the degree of symptom improvement and summarized in **TABLE 1**. Anterior decompression was the most common in 50 cases, and anterior discectomy fusion and anterior corpectomy fusion were introduced in 12 cases and 1 case, respectively. Posterior decompression and fusion were reported in 21 and 12 cases, respectively, and most were related to treatment of C1/2. Regarding the direct treatment of blood vessels, stenting, embolization, and bypass surgery were reported, and 6 cases, 3 cases, and 1 case were reported. Some symptoms remained in 8% of patients who underwent anterior decompression, and one case was reported in patients who underwent anterior corpectomy fusion and posterior fusion. In one report in which the vertebral artery was sacrificed during embolization, the symptoms remained the same as before surgery.

TABLE 1. Treatment method and prognosis

Surgery type	Anterior decom	ACDF	ACCF	Posterior decom	Posterior fusion	Stenting	Embolization	Bypass	Conservative treatment	Total
Number	50	12	1	21	12	6	3	1	12	118
Symptom improvement										118
NS	46	12	0	21	11	6	2	1	12	
PR	4	0	1	0	1	0	0	0	0	
TR	0	0	0	0	0	0	1	0	0	

ACDF: anterior cervical discectomy and fusion, ACCF: anterior cervical corpectomy and fusion, NS: no symptom, PR: partial remnant, TR: total remnant.

https://doi.org/10.13004/kjnt.2022.18.e63

DISCUSSION

BHS refers to a clinical condition characterized by symptomatic ischemia of the cerebellum or brainstem secondary to vertebral artery occlusion associated with multifactorial etiology.[16] The vertebral artery originates from the posterosuperior aspect of both subclavian arteries, runs medial to the anterior scalene and lateral to the longus colli muscle. The vessel most frequently enters the C6 transverse foramen (87%–89%); however, anatomical variations of entry at the C7 (3.5%–5.4%), C5 (6%), and above the C5 foramen (<1.4%) are observed in clinical practice.[4,10] The diameter of the transverse foramen increases from C3 to C6, and that at the C7 level is the smallest. The mean dimensions of all left-sided foramina at all levels are greater than those of the right side.[3,10]

When comparing anatomically, the cases reported mainly in C1 and C2 have been reported in previously reported studies.[16] However, a recently published review reported a higher probability of occurrence at the subaxial spine than at the atlas-axis level (52%).[15] Another review has also reported that this anomaly more frequently involved the subaxial spine (60%).[8] Etiologically, degenerative conditions such as osteophytosis were the most significant contributor (53%),[15] with the uncinate process being the most common site of osteophytosis. Approximately 50% of patients present with osteophytes that originate from the anteromedial portion, with consequent compression of the vertebral artery in a posterolateral direction. Reviews that have included >100 cases have reported that the lesions predominantly occurred on the left than on the right side and that this difference was statistically significant.[8,13] Notably, the number of patients with symptoms during rotation of the head to the right is greater than those with symptoms observed during head rotation to the left.[7,13] Interestingly, although symptoms appeared with head rotation to a particular side, the lesion could be located anywhere on either side.

Treatments for BHS include conservative management, stenting, decompression, and fusion surgery. Conservative treatment including neck collar usage, refraining from extreme neck rotations, and anticoagulant administration, among other such measures is preferred in patients with mild symptoms; symptoms tend to improve spontaneously, and patients may refuse surgery. Stenting is required in patients with symptoms secondary to atherosclerotic stenosis.[11]

Surgical treatment, which is recommended in most patients, is categorized into decompression or fusion surgeries. Decompression surgery is classified into laminectomy, transverse foramen opening, or resection of structures that cause occlusion. Approximately 8%–16% of patients had postoperative residual symptoms among those who underwent decompression surgery, and additional treatment was required in these cases.[8,15] However, most symptoms improved after fusion. Fusion is preferred in patients with multilevel disease, complex cases, or in those with pain radiation. Fusion performed concomitant with lesion removal minimizes movement of diseased vertebral levels and therefore prevents symptoms that occur during head rotation; therefore, the outcomes of fusion are better than those observed with decompression surgery. Many studies have reported no post-fusion recurrence.[8,13,15] Decompression surgery should be considered as the first-choice strategy because fusion is associated with permanent movement restrictions. However, symptoms may persist after decompression surgery, and a study has reported intraoperative angiography to overcome this limitation.[12] In the aforementioned study, the authors also recommended additional research regarding the usefulness of Doppler ultrasonography and indocyanine green videography. Nondominant vertebral artery compression can also be symptomatic and

https://doi.org/10.13004/kjnt.2022.18.e63

necessitates treatment.[7] Reportedly, surgical treatment is more effective than conservative management in patients with BHS.[13]

Syncope is a common symptom associated with multifactorial etiologies and is broadly classified into reflex-mediated syncope, orthostatic hypotension, and cardiovascular syncope.[9] Reflex-mediated syncope includes vagal and various types of situational syncope. Orthostatic hypotension refers to a reduction in blood pressure upon standing secondary to dehydration, drugs, or autonomic nervous system dysfunction. Cardiovascular syncope often results from arrhythmias. BHS is a rare condition that should however be considered in the differential diagnosis in patients with syncope.

Accurate diagnosis and prompt treatment are essential in patients with syncope to avoid accidents after loss of consciousness. BHS can be treated surgically; therefore, accurate diagnosis is important. Our patient presented with relatively clear symptoms that developed during head rotation in a single direction. BHS should be considered in the differential diagnosis in patients with recurrent syncope that is not clearly attributable to a specific etiology, particularly in patients in whom symptoms are associated with head turning. Dynamic angiography aids with diagnosis in such cases.

CONCLUSION

Syncope is a common symptom; however, syncope associated with rotational VBI is rare. Diagnosis is possible through cerebral angiography, and it is important to set up an appropriate treatment plan based on image. Notably, adequate decompression or fusion can relieve symptoms and improve patients' quality of life.

REFERENCES

1. Bukhari MK, Alghamdi SA. Ischemic stroke secondary to dynamic vertebral artery stenosis: case report and review of the literature. **Cureus 13**:e20167, 2021
PUBMED | CROSSREF
2. Bulsara KR, Velez DA, Villavicencio A. Rotational vertebral artery insufficiency resulting from cervical spondylosis: case report and review of the literature. **Surg Neurol 65**:625-627, 2006
PUBMED | CROSSREF
3. Cagnie B, Barbaix E, Vinck E, D'Herde K, Cambier D. Extrinsic risk factors for compromised blood flow in the vertebral artery: anatomical observations of the transverse foramina from C3 to C7. **Surg Radiol Anat 27**:312-316, 2005
PUBMED | CROSSREF
4. Cloud GC, Markus HS. Diagnosis and management of vertebral artery stenosis. **QJM 96**:27-54, 2003
PUBMED | CROSSREF
5. Ganzeboom KS, Mairuhu G, Reitsma JB, Linzer M, Wieling W, van Dijk N. Lifetime cumulative incidence of syncope in the general population: a study of 549 Dutch subjects aged 35-60 years. **J Cardiovasc Electrophysiol 17**:1172-1176, 2006
PUBMED | CROSSREF
6. Gould PA, Krahn AD, Klein GJ, Yee R, Skanes AC, Gula LJ. Investigating syncope: a review. **Curr Opin Cardiol 21**:34-41, 2006
PUBMED | CROSSREF
7. Iida Y, Murata H, Johkura K, Higashida T, Tanaka T, Tateishi K. Bow Hunter's syndrome by nondominant vertebral artery compression: a case report, literature review, and significance of downbeat nystagmus as the diagnostic clue. **World Neurosurg 111**:367-372, 2018
PUBMED | CROSSREF

https://doi.org/10.13004/kjnt.2022.18.e63

8. Jost GF, Dailey AT. Bow Hunter's syndrome revisited: 2 new cases and literature review of 124 cases. **Neurosurg Focus 38**:E7, 2015
PUBMED | CROSSREF
9. Kidd SK, Doughty C, Goldhaber SZ. Syncope (fainting). **Circulation 133**:e600-e602, 2016
PUBMED | CROSSREF
10. Lee V, Riles TS, Stableford J, Berguer R. Two case presentations and surgical management of Bow Hunter's syndrome associated with bony abnormalities of the C7 vertebra. **J Vasc Surg 53**:1381-1385, 2011
PUBMED | CROSSREF
11. Natello GW, Carroll CM, Katwal AB. Rotational vertebrobasilar ischemia due to vertebral artery dynamic stenoses complicated by an ostial atherosclerotic stenosis. **Vasc Med 14**:265-269, 2009
PUBMED | CROSSREF
12. Ng S, Boetto J, Favier V, Thouvenot E, Costalat V, Lonjon N. Bow Hunter's syndrome: Surgical vertebral artery decompression guided by dynamic intraoperative angiography. **World Neurosurg 118**:290-295, 2018
PUBMED | CROSSREF
13. Rastogi V, Rawls A, Moore O, Victorica B, Khan S, Saravanapavan P, et al. Rare etiology of Bow Hunter's syndrome and systematic review of literature. **J Vasc Interv Neurol 8**:7-16, 2015
PUBMED
14. Savage DD, Corwin L, McGee DL, Kannel WB, Wolf PA. Epidemiologic features of isolated syncope: the Framingham Study. **Stroke 16**:626-629, 1985
PUBMED | CROSSREF
15. Schulz R, Donoso R, Weissman K. Rotational vertebral artery occlusion ("bow hunter syndrome"). **Eur Spine J 30**:1440-1450, 2021
PUBMED | CROSSREF
16. Sorensen BF. Bow Hunter's stroke. **Neurosurgery 2**:259-261, 1978
PUBMED | CROSSREF
17. Strickland BA, Pham MH, Bakhsheshian J, Russin JJ, Mack WJ, Acosta FL. Bow Hunter's syndrome: surgical management (video) and review of the literature. **World Neurosurg 103**:953.e7-953.e12, 2017
PUBMED | CROSSREF
18. Tsutsumi S, Ito M, Yasumoto Y. Simultaneous bilateral vertebral artery occlusion in the lower cervical spine manifesting as Bow Hunter's syndrome. **Neurol Med Chir (Tokyo) 48**:90-94, 2008
PUBMED | CROSSREF
19. Velat GJ, Reavey-Cantwell JF, Ulm AJ, Lewis SB. Intraoperative dynamic angiography to detect resolution of Bow Hunter's syndrome: technical case report. **Surg Neurol 66**:420-423, 2006
PUBMED | CROSSREF
20. Vilela MD, Goodkin R, Lundin DA, Newell DW. Rotational vertebrobasilar ischemia: hemodynamic assessment and surgical treatment. **Neurosurgery 56**:36-43, 2005
PUBMED | CROSSREF

https://doi.org/10.13004/kjnt.2022.18.e63