Bow–Hunter Syndrome Due to Tandem Lesions; A Case Report

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Abstract

Keywords
► Bow–Hunter syndrome
► vertebral artery
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► osteophytes
► vertebral instability

Bow–Hunter syndrome (BHS) includes a form of stroke caused by dynamic factors affecting the extracranial vertebral artery (VA), such as rotation-induced flow disturbance at the atlantoaxial level. BHS is a rare condition that can result from a variety of factors, including abnormal bone compression and vertebral instability. So far, there have been no reports in which both vertebral instability and osteophytes compress the VA at different sites, leading to stroke. Herein, we present a rare case of stroke attributed to BHS involving thrombosis formation due to osteophyte compression at C3/4 and VA occlusion due to vertebral instability at the C4/5 overlapped. A 72-year-old male presented with rotational vertigo and vomiting associated with a cerebellar infarction. Digital subtraction angiography revealed severe stenosis of the left VA with an intra-arterial thrombus. Percutaneous transluminal angioplasty improved the stenosis; however, intravascular ultrasound revealed compression of the VA by osteophytes at C3/4. Additionally, left VA occlusion at C4/5 was confirmed because of vertebral instability. The osteophyte was removed via the anterolateral approach, and the VA was decompressed. The patient's postoperative course was uneventful, with no recurrence for more than 2 years. BHS can occur in the lower cervical spine when vertebral instability coincides with intimal arterial damage caused by osteophytes at other sites.

Introduction

Bow–Hunter syndrome (BHS) is a rare condition in which head rotation causes vertebral artery (VA) flow disturbance at the atlantoaxial level, leading to vertebrobasilar insufficiency.[1] Due to its anatomical course through the transverse foramen, the second se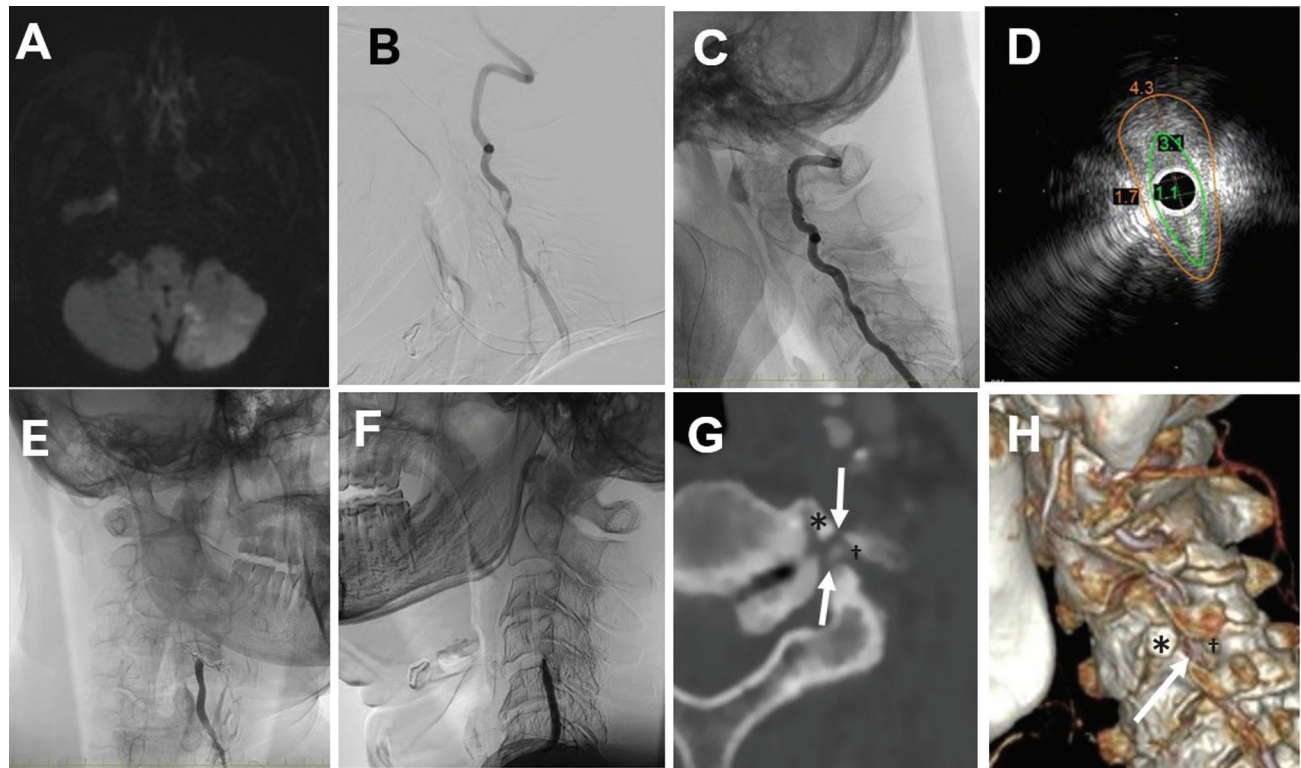gment of the VA can be compressed by abnormal bones such as osteophytes, leading to cerebral ischemia or embolism.[2–8] Additionally, the VA can be stenosed or occluded due to vertebral instability, further complicating the diagnosis and treatment. However, to our knowledge, there have been no reported cases in which vertebral instability and osteophytes separately compress the VA at different sites, leading to stroke.

Fig. 1 (A) Diffusion-weighted magnetic resonance imaging on admission showing an acute cerebellar infarction on the left side. (B) Cerebral angiography (CAG), lateral view, showing a severe stenosis at the vertebral artery (VA) with luminal thrombus at the C3/4 level. (C) CAG, neutral head position, post-percutaneous transluminal angioplasty (PTA), lateral view, showing improvement of the severe stenosis. (D) Intravascular ultrasound (IVUS) of the stenotic site (C3/4 level) of the left VA, post-PTA, showing that the site is surrounded with the calcified lesion. (E) CAG, left rotation of the head, post-PTA, anteroposterior (A-P) view, showing that the left VA was occluded at C4/5 level. (F) CAG, with head flexion, post-PTA, lateral view, left VA was occluded at the C4/5 level. (G) Computed tomography angiography (CTA), axial bone image at C3/4 level, showing that the stenotic site at VA was surrounded by abnormal bone structures (* and †). The white arrows indicate the stenotic left VA. (H) A reconstructed three-dimensional CTA showing that the VA is sandwiched by abnormal structures from the front (*) and back (†). The white arrow indicates that of the stenotic site (C3/4 level) of the left VA.

We encountered a rare case in which two abnormalities overlapped, leading to a stroke attributed to BHS. Specifically, these abnormalities included thrombosis formation due to intimal injury of the VA at the C3/4 osteophyte and cervical instability causing VA occlusion at C4/5.

Case Description

A 72-year-old male presented to our hospital with persistent rotational vertigo and vomiting upon awakening. He had a history of hypertension, hyperlipidemia, and left internal carotid artery stenosis, and was taking statins, aspirin, and antihypertensive medications. He had a smoking history of 20 to 40 cigarettes per day for > 30 years. On admission, his consciousness level was assessed as 15 on the Glasgow Coma Scale, with saccadic eye movements and horizontal gaze nystagmus. The patient presented with dysarthria, mild dysmetria, and mild decomposition. His National Institutes of Health Stroke Scale (NIHSS) score was 3, with left upper- and lower-limb ataxia and speech disorders. Magnetic resonance imaging revealed acute cerebellar infarction (►Fig. 1A).

Digital subtraction angiography (DSA) revealed severe stenosis with an intra-arterial thrombus in the left VA (►Fig. 1B). After 2 weeks of heparinization, percutaneous

transluminal angioplasty (PTA) was performed to treat the symptomatic left VA stenosis using Shiden 3.0 × 20 mm (Kaneka, Osaka, Japan) at 6 atm/2 minute and Genity 3.0 × 25 mm (Kaneka) at 6 atm/2 minute, resulting in stenosis improvement, with good blood flow in the neutral and right rotation head positions (►Fig. 1C). However, intravascular ultrasonography (IVUS) using the VOLCANO system (Volcano Japan Corp., Tokyo, Japan) and an IVUS catheter (Eagle Eye Platinum; Volcano Japan Corp.) indicated compression of the left VA by the osteophytes (►Fig. 1D). Furthermore, with the head in flexion and left rotation, left VA occlusion at the C4/5 was confirmed (►Fig. 1E, F). Considering the risk of stent breakage due to osteophytes, a stent was not placed, and the procedure was ended only when PTA was performed.

We diagnosed symptomatic VA stenosis occurring as a result of the simultaneous occurrence of two abnormalities: intimal damage due to compression at the left C3/4 osteophytes and VA occlusion due to vertebral instability of C4/5 (►Fig. 1G, H). The anterolateral approach was used to decompress the VA for osteophyte removal. The patient's head was rotated to the right while confirming the left VA blood flow with ultrasound (►Fig. 2A). Exposure of the osteophytes and the C3/4 anterior tubercle revealed compression of the VA (►Fig. 2B), and osteophytes were
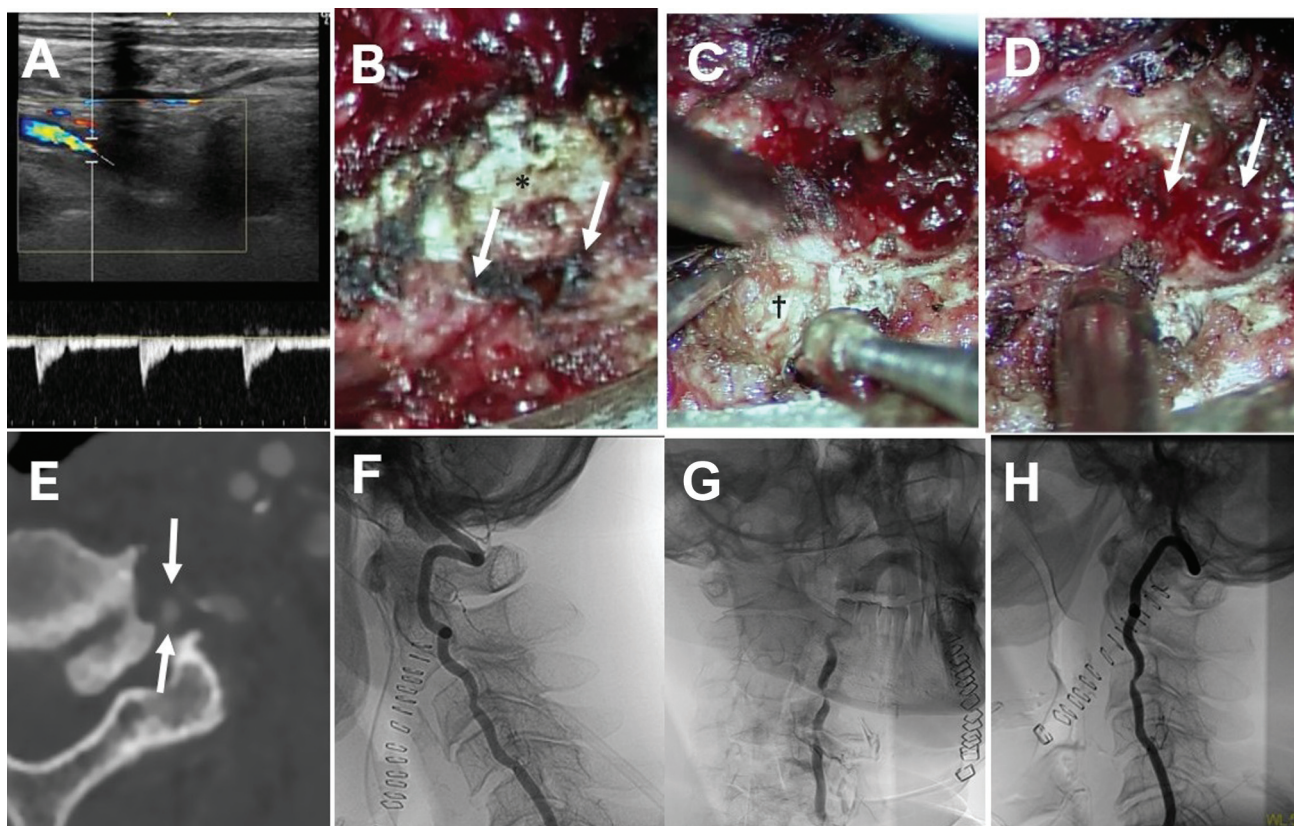
Fig. 2 (A) The intraoperative ultrasound flow study of the patient's left vertebral artery (VA) before operative procedure; the observation of blood flow in the VA during the diastolic period confirmed that the VA was not obstructed. (B) Intraoperative photograph showings that the abnormal osteophytes which present in front of the VA (*) are compressing the artery (white arrows). (C) Intraoperative photograph showing that the VA was transported anteriorly, while the osteophyte (†) which is located posterior to the VA was drilled. (D) Intraoperative photograph showing that the VA (white arrows) was completely decompressed. (E) Postoperative computed tomography angiography (CTA), axial image, bone image of C3/4 level, showing effective decompression of the VA (paired arrows) due to the removal of surrounding abnormal osteophytes. (F) Cerebral angiography (CAG), postdecompression procedure, neutral head position, showing good blood flow in the left vertebral artery (VA) at the C3/4 and C4/5 levels. (G) CAG, left rotation of the head, postdecompression, showing that the VA was still stenotic and the blood flow was delayed, but with no arterial occlusion. (H) CAG, flex of the head, postdecompression, showing no occlusion of the VA.

removed. In addition, the base of the C3/4 anterior tubercle was drilled to expose the VA. Further, we removed the hypertrophic osteophyte that arose from the intervertebral foramen of C4, as it compressed the VA from behind (►**Fig. 2C**). Finally, the relief of tension on the VA was confirmed (►**Fig. 2D**). Postoperatively, computed tomography angiography confirmed the removal of the C3/4 osteophyte and complete decompression of the VA (►**Fig. 2E**).

DSA obtained on the 7th postoperative day revealed no significant stenosis at C3/4 or C4/5 in the neutral position (►**Fig. 2F**). However, residual severe stenosis of the left VA with delayed flow in left head rotation was observed (►**Fig. 2G**). Minimal stenosis at C3/4 was observed in head flexion compared with the neutral position, but no VA occlusion occurred at C4/5 (►**Fig. 2H**). The patient was discharged on postoperative day 10 with a modified Rankin Scale score of 0 and an NIHSS score of 0. The patient has remained recurrence-free for 2 years.

Discussion

Herein, we presented a case of BHS in which the left VA was compressed by osteophytes between the C3 and C4 interver-

tebral foramen, causing intimal damage and stenosis of the VA. Additionally, vertebral instability at the C4/5 level was resulting in an abnormal VA course, causing occlusion of the left VA during left rotation or extension of the head. These abnormalities caused thrombosis at the C3/4 level, leading to cerebellar infarction when the thrombus was scattered. In the present case, we aimed to prevent recurrent stroke by removing the obvious thrombotic causes of the osteophytes. Although the lesions occurred at separate sites at the C3/4 and C4/5 levels due to overlapping causes of osteophyte and vertebral instability, resection of the osteophyte led to a favorable outcome.

This case report has several clinical implications. First, this experience shows that VA cryptogenic occlusion due to vertebral instability, VA latent stenosis, and/or intimal damage caused by osteophytes at different sites can cause cerebellar infarction when these two events occur simultaneously. To the best of our knowledge, no similar cases have been reported thus far.

In the present case, we determined that the osteophytes at the C3/4 directly caused compression of the VA and led to occlusion during rotation. Furthermore, we inferred from preoperative DSA imaging that the presence of vertebral

instability caused occlusion of the VA at C4/5 during extension. Additionally, we hypothesized that the formation of these abnormal osteophytes demonstrates vertebral instability.

There are no specific treatment guidelines for BHS, which leads to difficulty in determining the treatment approach. Approximately half of all patients treated with antiplatelet agents experience recurrence within several months to years, suggesting that some form of surgical intervention is recommended.[9] In cases presenting with abnormalities at two sites, such as ours, it is essential to clearly understand the pathogenesis of cerebellar infarction as two separate entities. Although most individuals with vertebral instability remain asymptomatic due to dynamic effects, the addition of other pathological factors can lead to cerebellar infarction. Therefore, personalized treatment approaches are required. In our case, we aimed to remove the pathological factor of intimal damage by decompressing the osteophyte, thereby limiting the impact to only dynamic effects.

In addition, BHS can occur at the lower cervical spine, although it is typically associated with VA occlusion of the upper cervical spine (C1/2) during head rotation. Reports of BHS in the lower cervical spine are rare; however, when they do occur, they are often associated with osteophytes.[2,4,5,10–13] Prior case reports of vertebral instability have shown that the upper vertebral body on the affected intervertebral segment slides backward on the side where it has rotated, while the VA in the transverse foramen is subjected to overstretching, leading to compression of the VA. Furthermore, in the presence of osteophytes, the VA is more strongly compressed and prone to occlusion.[3] Therefore, when a BHS is found at the lower cervical spine, it is necessary to actively check for vertebral instability to make an accurate diagnosis and decide on an appropriate treatment strategy.

Finally, IVUS can provide clues for the diagnosis of BHS. In this case, we used IVUS to confirm the presence or absence of VA dissection or thrombus, the measurement of the vessel diameter, and the nature of the VA stenosis, finding that the osteophyte surrounding the VA caused arterial compression. To the best of our knowledge, which is similar to our case, only one reported case has shown that IVUS is useful for diagnosing BHS.[14]

Conclusion

BHS may occur in the lower cervical spine. Further, this phenomenon can be caused by two coinciding factors: vertebral instability and intimal damage due to osteophyte at a different site.

Note
At the 53rd Annual Meeting of the Japanese Society on Surgery for Cerebral Stroke on March 9, 2024.

Authors' Contributions
All authors contributed to the conception and design of the study. Material preparation and data collection were performed by all the authors. Data analysis was performed by S.Y. and J.N,. The first draft of the manuscript was written by S.Y., and all authors commented on the previous versions of the manuscript. All the authors have read and approved the final manuscript.

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