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CASE REPORT



Bow hunter's syndrome after cervical laminoplasty in a patient with rheumatoid arthritis with bony ankylosis in the cervical spine: a case report

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ABSTRACT

Bow hunter's syndrome, or rotational vertebral artery (VA) occlusion, refers to vertebrobasilar insufficiency due to mechanical occlusion of the VA. We present a case of surgical treatment for bow hunter's syndrome that occurred after cervical laminoplasty in a patient with rheumatoid arthritis with bony ankylosis of the facet joints. A 59-year-old female with rheumatoid arthritis experienced sudden incomplete left hemiplegia. Fifteen months earlier, the patient had undergone cervical decompression surgery between C3 and C7. MRI of the head showed cerebral infarction in the right VA area, while vertebral angiography with the head rotated to the right revealed that the right VA was occluded at the level of C3–C4. The patient was successfully treated via posterior cervical fusion from C2 to C7. Patients with rheumatoid arthritis have a potential risk of cervical bony ankyloses. Cervical laminoplasty for patients with cervical bony ankyloses can induce rotational VA occlusion due to spinal rotational instability.

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postoperative spinal
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Introduction

Bow hunter's syndrome (BHS), also known as rotational vertebral artery (VA) occlusion, is a form of vertebrobasilar insufficiency due to mechanical occlusion of the VA at the atlantoaxial or subaxial level. The most common symptoms of BHS include dizziness, vertigo, syncope, dysarthria, nausea and dysphagia [1–3]. At the subaxial level, the flow of the VA is obstructed during head rotation due to osteophytes, the longus colli muscle or degenerative cervical spondylosis [3–7].

Rheumatoid arthritis (RA) often causes destruction and instability of the cervical spine, such as atlantoaxial subluxation, vertical subluxation or subaxial subluxation. Patients with RA often present with bony ankyloses in the cervical spine, as well as the hand and wrist joints [8–10]. Bony ankylosis of the facet joints in the cervical spine has been demonstrated in patients with severe RA, and this bony ankylosis tends to induce adjacent segment pathology [9]. However, little is known about the influence of bony ankyloses on the cervical spine after cervical decompression surgery. We present a case of surgical treatment for BHS after cervical laminoplasty in a patient with RA with cervical bony ankyloses. Written consent was obtained from the patient for publication of the data.

Case presentation

A 59-year-old female presented with sudden incomplete left hemiplegia, and was delivered to another hospital by emergency transport. During the night before the onset of symptoms, the patient had slept with her head rotated to the right, and subsequently woke up with left hemiplegia. She had no history of head rotation causing symptoms of vertebrobasilar insufficiency such as dizziness, vertigo, syncope, dysarthria, nausea and dysphagia. In the 15 months before presentation, the patient had undergone cervical laminectomy at C3 and laminoplasty from C4 to C7. She had been diagnosed with RA 36 years earlier, and had been treated with prednisolone, salazosulfapyridine, leflunomide and golimumab (Steinbrocker stage IV, functional class IV).

MRI of the head showed infarction in the right cerebellar and pons region, which is the area that the VA supplies with blood (Figure 1). Blood analysis showed no evidence of coagulation and autoimmune abnormalities, and cardiac valve disease or a patent foramen ovale was not identified by transoesophageal echocardiography. Ultrasonography of the right VA showed a marked decrease in the flow velocity with the head rotated to the right, while ultrasonography of the left VA showed a marked decrease in the flow velocity with the head rotated

to the left. Bony ankylosis at the bilateral atlantoaxial joints and bilateral C4–C5 facet joints was visualized on parasagittal CT before cervical laminoplasty (Figure 2). Vertebral angiography revealed that the right VA was occluded at the C3–C4 level when the head was turned to the right, while the left VA was occluded at the C3–C4 level when the head was turned to the left (Figure 3(B,D,E)). In neutral position, there was no occlusion or constriction of the bilateral VA, and there was adequate blood supply to the posterior circulation (Figure 3(A,C)). At about 1 year after laminoplasty, the retrospondylolisthesis at C3 had worsened and osteosclerotic changes had appeared at the C3–C4 vertebral endplates (Figure 4). Cervical MRI showed no evidence of spinal cord compression (Figure 5). Based on these findings, thrombus formation in the right vertebral artery may be occurred by decreased blood flow during the night because of her head rotation to the right. And cerebral embolism in the vertebral artery region developed when occlusion of the vertebral artery was released.

Heparinization was performed on admission day, and posterior cervical fusion was performed between C2 and C7 one month later (Figure 5). The

postoperative course was uneventful, and the patient has not had further cerebral infarction. At 1 year after the onset of symptoms, the left hemiplegia had improved with conservative therapy and the patient was able to walk with a cane. Parasagittal CT performed 1 year postoperatively confirmed bony fusion from C2 to C7.

Discussion

The clinical course of the present patient suggests two important clinical issues. First, cervical laminoplasty for patients with cervical bony ankyloses can lead to rotational spinal instability. Second, this spinal rotational instability can then cause subaxial rotational VA occlusion.

Recent retrospective reviews of BHS have included more than 120 reported cases [5,11]. Some reports showed that the atlantoaxial joint is the most common level of VA occlusion [11,12], while one literature review reported that occlusion was most commonly located in the mid-lower cervical spine [5]. In subaxial BHS, the VA is often occluded by osteophytes and disc herniation [4].

The mechanism of VA insufficiency in patients with RA can be due to a number of causes: atlantoaxial instability and subluxation [13–17], displacement of the odontoid process into the foramen magnum [18] and destructive changes at the occipitocervical junction [16]. In the present case, rotational occlusion of the VA occurred at the C3 level after cervical laminoplasty. So far, there have only been two reported cases of secondary BHS after cervical surgery [4,19]. In these cases, rotational vertebral instability occurred as adjacent segment disease after cervical fusion. Therefore, the present case is unique in that the symptoms occurred after cervical decompression without fusion.

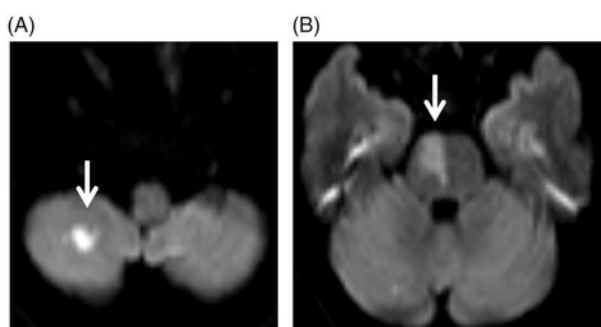


Figure 1. Axial diffusion-weighted magnetic resonance imagery of the brain at the onset of symptoms shows high-intensity areas in the right cerebellum (A) and pons (B).

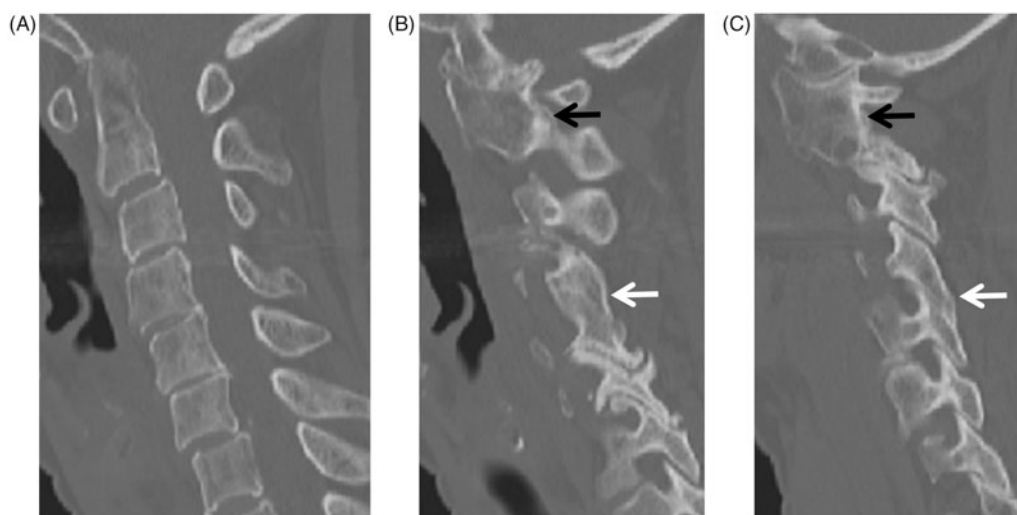


Figure 2. Cervical CT before the initial surgery. Mid-sagittal (A), right parasagittal (B) and left parasagittal (C) views. There were bony ankyloses of the bilateral atlantoaxial joints (black arrows) and bilateral C4–C5 facet joints (white arrows).

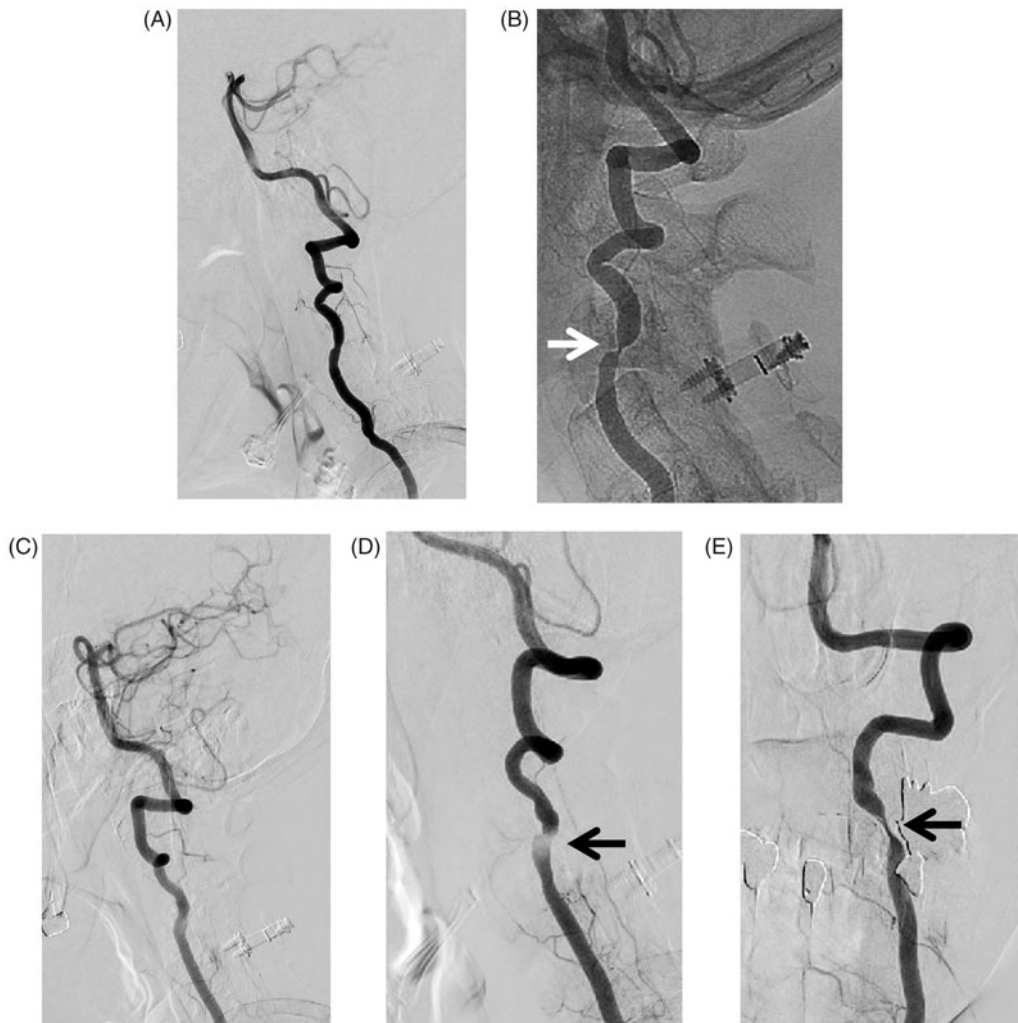


Figure 3. Lateral view of right vertebral angiography in the neutral position (A), and with the head rotated to the right (B). Lateral view of left vertebral angiography in the neutral position (C). Left vertebral angiography with the head rotated to the left in lateral view (D), and anteroposterior view (E). The vertebral artery was occluded at the C3–C4 level with the head rotated to the right (white arrows), and the left vertebral artery was occluded at the C3–C4 level with the head rotated to the left (black arrows).

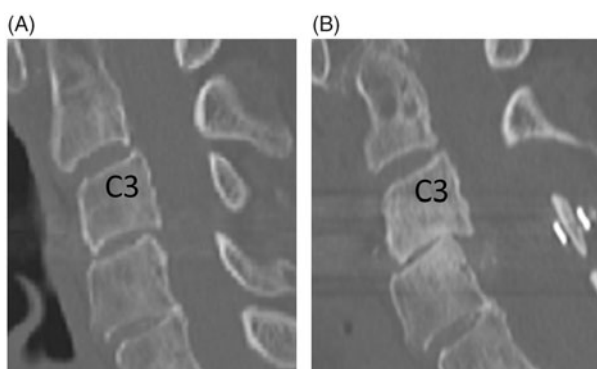


Figure 4. Midsagittal cervical CT before laminoplasty (A) and at 1 year after laminoplasty (B).

Cervical laminoplasty for patients with cervical bony ankyloses can lead to rotational spinal instability. RA often affects the cervical spine, as well as the hand and wrist joints, and the prevalence of bony ankyloses of the cervical spine is reportedly 9–26% [8–10]. Biomechanically, the atlantoaxial joint is the most rotatable joint in the cervical spine, with a normal range of rotation of 40–45° [20,21]. The present

patient had bony ankyloses at the atlantoaxial joint and the C4–C5 facet joints before laminoplasty, and the C3 vertebral posterior slippage deteriorated and osteosclerosis appeared at the C3–C4 vertebrae after laminoplasty. Therefore, rotational mechanical stress on the C3 vertebra, which was the segment adjacent to the ankylosed joints, may increase after cervical laminectomy at C3 with C4–C7 laminoplasty.

There are several treatments for BHS, including conservative therapy using a neck brace to avoid neck rotation [22]. Literature review of BHS [5] reported that 107 of 116 patients underwent surgical intervention. Nine cases were initially managed conservatively, including 4 cases in which patients chose conservative therapy despite recommended surgery. Two cases initially treated with neck brace and subsequently underwent surgery. Therefore, surgical intervention was recommended in patients with BHS. In surgical treatment for BHS, anterior and posterior procedures, with or without decompression and with or without fusion, have

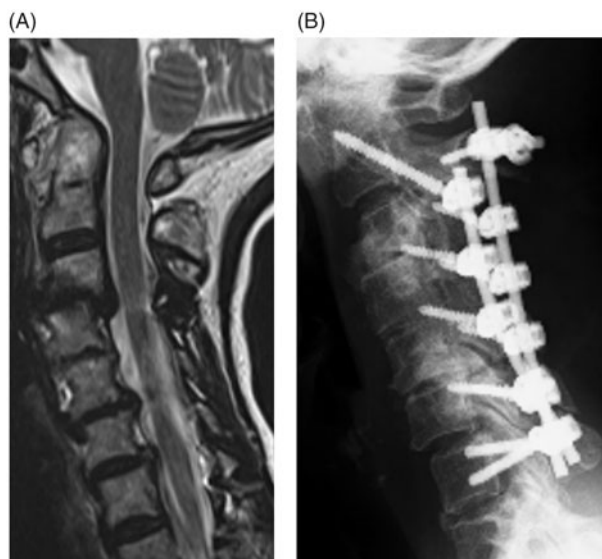


Figure 5. Midsagittal T2-weighted magnetic resonance image at 1 year after cervical laminoplasty (A) and lateral cervical spine radiograph after posterior cervical fusion (B).

been used. Based on the pathology of BHS, cervical fusion of the affected segment without decompression has been proposed [23]. Our patient was treated with posterior fusion due to rotational instability that caused cerebellar and pons infarction, and the treatment successfully prevented the occurrence of further neurological symptoms.

In conclusion, the potential risk of cervical bony ankyloses in patients with RA should be recognized. Cervical laminoplasty for patients with cervical bony ankyloses can induce rotational VA occlusion due to spinal rotational instability.

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Ethical Approval

Not Applicable.

Conflict of interest

None.

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