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Bow hunter's syndrome: temporary obstruction of blood flow in the affected vertebral artery during posterior occipitocervical fusion. Illustrative case

Takeru Yokota, MD,[1] Koji Otani, MD, DMSc,[1] Junichi Handa, MD, PhD,[1] Takuya Nikaido, MD, PhD,[1] Takao Kojima, MD, PhD,[2] Naoki Sato, MD, PhD,[3] and Shinichi Konno, MD, PhD[1]

Departments of [1]Orthopaedic Surgery and [2]Neurosurgery, Fukushima Medical University School of Medicine, Fukushima City, Fukushima Prefecture, Japan; and [3]Department of Neurosurgery, Masu Memorial Hospital, Nihonmatsu City, Fukushima Prefecture, Japan

BACKGROUND Bow hunter's syndrome (BHS) is a rare condition characterized by mechanical impingement of a vertebral artery (VA) during neck rotation followed by vertebrobasilar insufficiency. Posterior fusion is a typical surgical method for BHS.

OBSERVATIONS The case of a 70-year-old Japanese man who presented with presyncope that occurred during right cervical rotation is reported. Given the possibility of vertebrobasilar insufficiency, digital subtraction angiography and computed tomography angiography were performed and showed a hypoplastic right VA and severe stenosis of the left VA over the occiput (O)–C2 level. The blood flow of the left VA was interrupted by right cervical rotation, with resumption of blood flow on left cervical rotation. BHS was diagnosed, and posterior fusion at the O–C2 level was performed. Immediately after implant fixation, selective arteriography confirmed disruption of blood flow in the left VA. The rods were removed immediately; resumption of blood flow was confirmed; and the rods were refixed, anatomically bent with slight left cervical rotation. Then, sustained blood flow in the left VA was confirmed.

LESSONS Posterior fixation for BHS can induce VA occlusion due to minor changes in cervical spine alignment. Intraoperative selective arteriography is a necessary tool to identify occlusion of the affected VA.

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KEYWORDS bow hunter's syndrome; occipitocervical plate; posterior fusion; selective angiography

Bow hunter's syndrome (BHS), also known as rotational vertebral artery (VA) occlusion syndrome, was reported by Sorensen in 1978.[1] In BHS, the neurological symptoms of basilar artery circulatory insufficiency, including syncope and dizziness, occur due to physical VA stenosis associated with cervical rotation. There are still few reports of BHS, and no standard treatment has been established. It has been reported that conservative treatment is effective in only 37%, and its effectiveness is limited.[2] Decompression and/or fusion has been reported as typical surgical treatment for BHS.[3] In recent years, endovascular treatment has also been reported.[3]

With such surgical treatment, VA obstruction is one of the most severe complications.[4] On one hand, the decompression maneuver involves the risk of vascular injury, because the tissue surrounding the narrowed VA must be manipulated. On the other hand, in spinal fusion, vascular injury may occur when inserting a screw. Therefore, in surgical treatment for BHS, it is necessary to pay attention to VA damage. However, to the best of our knowledge, there has been no report of BHS in which the affected VA was obstructed during spinal fusion using an occipitocervical (OC) plate.

In the present case, interruption of affected VA blood flow was identified using intraoperative selective angiography, and it was then successfully restored. In patients with BHS, intraoperative evaluation of blood flow may be useful to detect and prevent VA occlusion.

Illustrative Case

History and Presentation

A 70-year-old man had a left cerebral infarction and underwent left carotid endarterectomy. Two months after the operation, he had an

ABBREVIATIONS BHS = bow hunter's syndrome; CT = computed tomography; O = occiput; OC = occipitocervical; VA = vertebral artery.

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articulation disorder, left hemiplegia, and right conjugate deviation, and he was diagnosed with a left cerebellar infarction. Selective angiography showed that the V3 segment of the left VA over the occiput (O)–C2 level had severe stenosis, blood flow was interrupted by right rotation of the neck, and blood flow was restored by left rotation. The examination showed that the right VA was completely occluded, and collateral circulation visualized the vertebral-basilar artery system. BHS was suspected, and he was referred to our department. His past history included hypertension, type 2 diabetes mellitus, heart failure, and laminoplasty (C2–7) for cervical spondylosis at the age of 62 years. He was receiving various drugs, including clopidogrel (75 mg/day) and aspirin (100 mg/day).

The patient's main complaint was dizziness induced by right rotation of the neck. He was wearing a soft neck collar and was able to walk without a cane. Physical findings showed muscle weakness on the left side of the body. Manual muscle testing of the left upper limb was 3–4 and was 4 in the left lower limb. He had no sensory disturbance, and deep tendon reflexes, as well as Hoffmann's and Wartenberg's reflexes, were enhanced.

Imaging Studies

Radiographs of the cervical spine showed spondylotic changes between each vertebra and postlaminoplasty status at the C2–7 levels. Functional flexion/extension radiographs of the cervical spine did not show instability. Computed tomography (CT) of the cervical spine showed a defect in the posterior arch of the atlas, which was considered to be a congenital anatomical abnormality at the OC junction, and the posterior arch of the axis and the occipital bone were close to each other (Fig. 1). On contrast-enhanced CT, blood flow in the right VA was completely disrupted, and the left VA had severe stenosis in the V3 segment between the posterior arch of the axis and the occipital bone (Fig. 2). On selective angiography of the V3 segment of the left VA, blood flow was interrupted by right rotation of the neck and was restored by left rotation (Fig. 3).

On the basis of the above clinical course and imaging findings, BHS was diagnosed, and surgery was planned. Because the affected VA had severe stenosis, decompression around the narrow segment was thought to have a high risk of VA injury, and posterior OC fusion over O–C2 was planned. To evaluate intraoperative blood flow of the affected VA, selective angiography during the operation was planned.

Two days before O–C2 fusion, temporary fixation with a halo-vest was performed with the patient conscious. The halo-vest was placed in the neutral position of the neck under local anesthetic. After fixation of the halo-vest orthosis, it was confirmed that there were no complications, such as interruption of blood flow in the affected VA or the appearance of dysphagia.

Operation and Postoperative Course

The operation was performed with the patient in the prone position and under general anesthesia, and a multidimensional surgical imaging system (O-arm, Medtronic) was used. The halo-vest was removed immediately after the patient's position was changed to the prone position. A catheter was placed just before the start of surgery to enable intraoperative angiographic diagnosis as needed, selective angiography was performed to confirm blood flow of the left VA (Fig. 4A), and the operation was started. The approach was a posterior approach, expanding the range from the occipital bone to the C3 spinous process. A reference frame for the O-arm was placed on the C3 spinous process, and subsequent operations were performed under O-arm guidance. First, the C2 screw was inserted. Next, an occipital plate (Medtronic Sofa Moadaneck VERTEX OC plate) was placed on the external occipital protuberance. The occipital bone plate and C2 screw were connected by rods. The shape of the prebent rods seemed to be well fitted against the plate and screw, and it was judged that the correction force applied to the lesion site was not large; therefore, no further bends were applied to the rods initially.

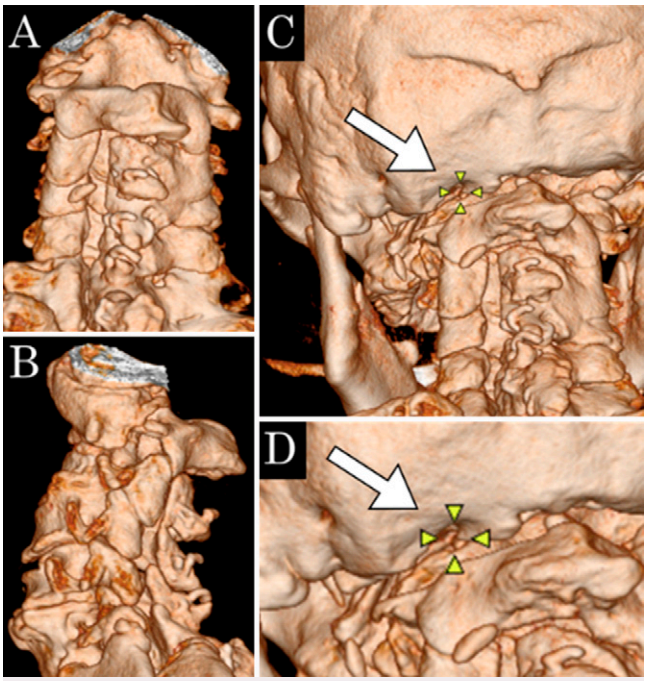

FIG. 1. Three-dimensional CT scans: posterior (**A**) and lateral (**B**) views. The *white arrows* show where the atlas and occipital bone are close together (**C and D**).

When intraoperative angiography was performed after the fixation, the left VA was slowly imaged up to the atlas but not beyond the atlas (Fig. 4B, Video 1). The rods were immediately removed, and angiography was repeated, confirming that the blood flow in the left VA was restored to the same as before the operation (Fig. 4C). The time from the interruption of blood flow to its resumption was about 20 minutes. It was considered that the correction of implant fixation caused the alignment change, and compression of the left VA by the surroundings resulted in the interruption of blood flow. The fixation angle of the cervical spine was changed from neutral to a left rotation position of less than 5 degrees, and the shape of the rod was bent carefully and anatomically so that the correction of the rod worked as little as possible. The rods were refixed, and a connector was fixed to the rods around the level of the C2 screw. Subsequent angiography showed that blood flow was maintained at the same level as before (Fig. 4D). Finally, a half-layer of bone was collected from the left iliac bone and fashioned into an elongated bone, which was transplanted into the decorticated C2 vertebral arch and occipital bone (Fig. 5).

VIDEO 1. Clip showing intraoperative selective arteriography when vertebral artery occlusion was first detected. Click here to view.

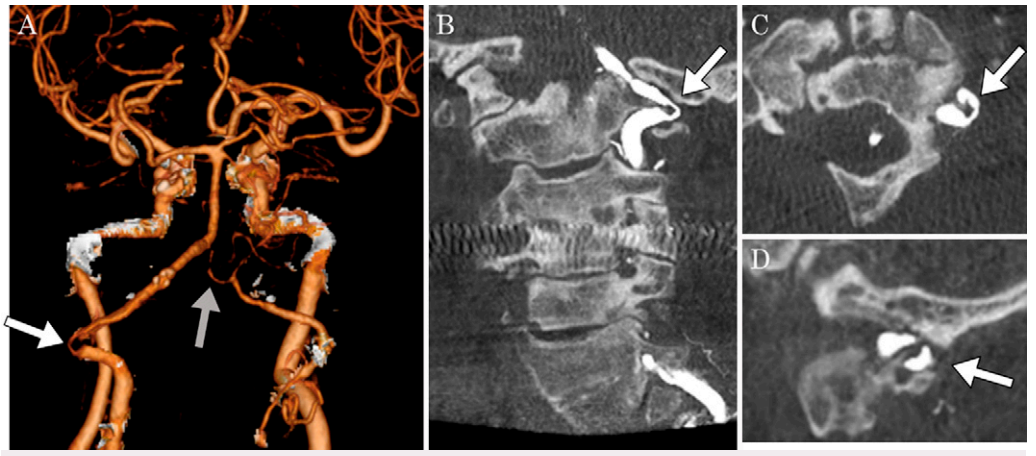

FIG. 2. A: Three-dimensional contrast-enhanced reconstruction of brain blood vessels. **B**–**D:** Slices of the lesion in coronal (**B**), horizontal (**C**), and sagittal sections (**D**), respectively. The *white arrows* show that the left vertebral artery is kinked in the region where the atlas and occipital bone are in close proximity. The *gray arrow* indicates the area of absent contrast in the right vertebral artery.

There were no postoperative complications from the end of surgery to the time of discharge. Contrast-enhanced CT immediately after surgery and 8 days after surgery showed no disruption of blood flow in the left VA. The patient wore a soft neck collar while in the hospital and was discharged to another hospital 11 days after the operation.

At outpatient follow-up, 6 months after the operation, the cervical spine was capable of active rotation of 5 degrees to the right and 10 degrees to the left. The dizziness induced by right rotation movement disappeared completely after the operation, and no new neurological symptoms appeared in the follow-up period up to 6 months after the operation.

Discussion

Observations

In the present case, OC fusion was performed for BHS. Intraoperative disruption of blood flow in the VA was identified by selective angiography. By forming the rods anatomically to reduce the correction, it was possible to prevent reinterruption of blood flow after fixation.

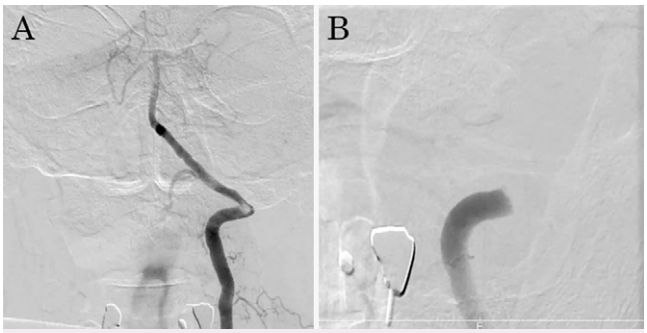

FIG. 3. On selective arteriography, the left vertebral artery is contrasted in the midcervical position (**A**), but there is contrast interruption in the V3 segment of the left vertebral artery on right cervical rotation (**B**).

Disruption of VA blood flow was found after implant fixation by performing intraoperative angiography, and it was possible to restore blood flow as soon as possible. There are some reports of the importance of intraoperative arteriography during treatment for BHS.[5–10] For example, intraoperative VA angiography is useful for evaluating arterial injury in screw fixation or for evaluating whether decompression was properly performed in decompression. In the present case, posterior fusion between the cervical spine and

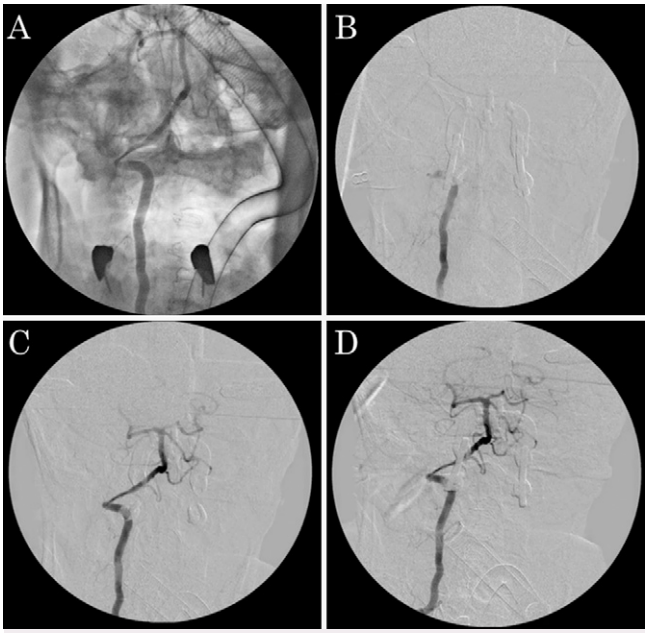

FIG. 4. On intraoperative selective arteriography, there is no occlusion of the left vertebral artery immediately before surgery (**A**); the left vertebral artery is disrupted in the V3 segment after the rod is fixed (**B**); the occlusion of the left vertebral artery is released immediately after the rod is removed (**C**); and no reocclusion of the left vertebral artery occurs after the rod is refixed (**D**).

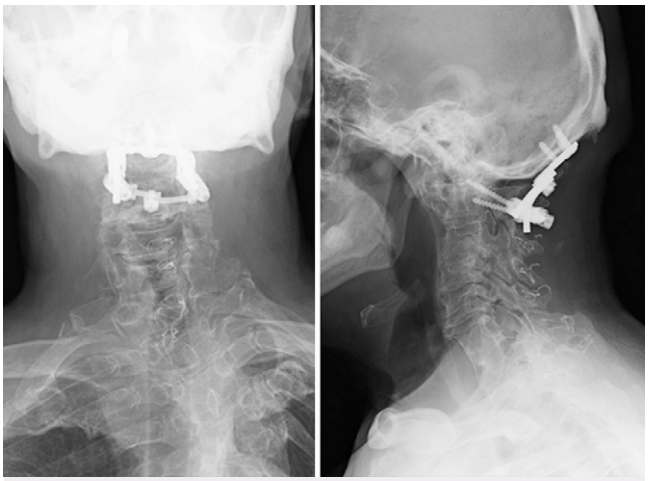

FIG. 5. Postoperative radiographs.

occipital bone was performed. Though the screw and plate fixation procedure did not cause vascular injury, it is thought that the subsequent rod fastening caused dislocation of the OC junction and that left VA blood flow was interrupted. If the blood flow of the left VA had not resumed, there could have been a significant impact on functional prognosis and life prognosis. However, because the blood flow was restored as soon as possible, no neurological complications occurred after the operation. From our experience, we insist that intraoperative arteriography is an essential examination to prevent VA occlusion due to the surgical procedure in BHS.

In the present case, the affected VA appeared to have been narrowed by the dislocation of the OC junction due to the fastening of the rods, resulting in blood flow interruption. Various complications caused by posterior fusion have been reported, and one of the typical complications is VA injury caused by implant fixation.[11] However, to the best of our knowledge, there have been no reports of impaired blood flow in the VAs after the rod-fastening operation, as observed in the present case. The OC plate is an effective implant that improves the malalignment and instability of the OC junction. In other words, use of the same implant can result in local alignment changes at the OC junction.[12] In the present case, halo-vest fixation was performed to maintain the most appropriate head position before surgery during fusion. However, past reports have shown that even with preoperative halo-vest fixation, the corrective force of the OC plate can change the anatomical position of the OC junction. In the present case, the alignment of the OC junction appeared to have been changed by the correction due to rod fastening. The left VA was physically compressed, and the blood flow was interrupted. During posterior fusion with an OC plate, it should be noted that changes in the alignment of the OC junction can also damage the affected VA.

Intraoperative reflection points in this case included the implant fixation procedure and the duration of blood flow interruption in the VA. First, as mentioned above, there is a risk of disruption of blood flow in the VAs due to slight alignment changes in the cervical spine. Therefore, when fixing the implant, it was considered important to form the rod so that the alignment before and after fixing did not change. In the present case, no new neurological symptoms appeared after the operation, but the time to restore blood flow was relatively long at 20 minutes. Originally, a neurosurgeon was scheduled to perform vertebral angiography immediately after the initial fixation, but due to overlap of emergency surgery in the same department, the start of selective arteriography in this case was delayed. Because the delay in resuming blood flow could have led to death, as well as serious neurological complications, in cases such as the present one, a better way to deal with this is needed. As described above, in BHS surgery, various factors can affect VA blood flow, so it is necessary to pay close attention to the preparation for surgery and to the procedure during surgery.

Lessons

A case of temporary occlusion of blood flow in the affected VA after posterior OC fusion for BHS is reported. In BHS, blood flow in the affected VA may be disrupted due to minor changes in cervical alignment caused by implant fixation. Therefore, it is important to evaluate blood flow during surgery by selective angiography.

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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: Otani, Yokota. Acquisition of data: Yokota, Nikaido, Sato. Analysis and interpretation of data: Yokota. Drafting the article: Otani, Yokota, Sato. Critically revising the article: Kojima. Reviewed submitted version of manuscript: Otani, Nikaido, Kojima, Konno. Approved the final version of the manuscript on behalf of all authors: Otani. Administrative/technical/material support: Handa.

Supplemental Information

Video

Video 1. https://vimeo.com/764575568.

Correspondence

Koji Otani: Fukushima Medical University School of Medicine, Fukushima City, Fukushima Prefecture, Japan. kotani@fmu.ac.jp.