Doppler Ultrasonography and CT Angiography Demonstrate Positional Occlusion of Vertebral Artery Associated with One-sided Destruction of the Atlantoaxial Lateral Mass Caused by Rheumatoid Arthritis: A Case Report

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Abstract

Study Design. Case report of a patient with rheumatoid arthritis (RA) and a positional occlusion of the left vertebral artery (VA).

Objective. To describe the utility of Doppler ultrasonography and computed tomography (CT) angiography for the diagnosis of positional VA occlusion.

Summary of Background Data. In previous reports of positional VA occlusion in RA, angiography has been used for the diagnosis. However, it is difficult to demonstrate the three-dimensional relationship between the arteries and the bone structure with angiography.

Methods. An 83-year-old man with a 20-year history of RA complained of severe vertigo when he leaned his head in the left-anterior direction. CT angiography in the neutral position revealed that the left VA was pinched between the posterior rim of the transverse foramen of C1 and the transverse process of C2. Doppler ultrasonography demonstrated positional VA occlusion and a severe reduction in blood flow at the position that most readily induces vertigo. Because the space between the transverse foramens of left C1 and C2 was reduced with the destruction of the left C1/C2 lateral masses, slight rotation and anterior shift of C1 led to the occlusion of the VA.

Results. After posterior O–C2 fusion at the reduced position, the VA occlusion and vertigo disappeared.

Conclusion. Doppler ultrasonography and CT angiography allow valuable measurements in the diagnosis of positional VA occlusion. The one-sided destruction of the C1/2 lateral masses might be a causal factor for VA occlusion in RA. This is the first report of a new pathomechanism underlying positional VA occlusion demonstrated with three-dimensional

CT angiography.

Key Points

- The one-sided destruction of the C1/2 lateral masses might be a causal factor for VA occlusion in RA.
- Doppler ultrasonography can show positional changes in the blood flow of VA.
- Three-dimensional CT angiography can clearly demonstrate the three-dimensional relationship between VA occlusion and the bone structure.
- The combination of ultrasonography and CT angiography is less invasive and more informative than angiography

53

54 **Mini Abstract**

55 An RA patient complained of severe vertigo when he leaned his head in the left-anterior
56 direction. CT angiography revealed occlusion of left VA between the posterior rim of the
57 transverse foramen of C1 and the transverse process of C2. After posterior O–C2 fusion, the
58 VA occlusion and vertigo disappeared.

Introduction

Patients with rheumatoid arthritis (RA) suffer the destruction of multiple joints, including the cervical spine. Atlantoaxial instability (AAI) is the most common cervical lesion of RA. The vertebral artery (VA) runs through the transverse foramens of C6–C1 and into the dura in the occipital region. Therefore, cervical lesions such as cervical spondylosis or AAI have been reported to lead to the occlusion or stenosis of the VA.[1,2]

In most reports of VA in patients with RA, angiography has been used in the diagnosis of positional VA occlusion.[3-12] Although angiography is suitable for the detection of positional VA occlusion, it is invasive and ineffective in its presentation of the three-dimensional relationship between the arteries and the bone structure.

In this case report of a patient with RA, Doppler ultrasonography and computed tomography (CT) angiography demonstrated the positional occlusion of the left VA at C1/C2, attributed to the one-sided destruction of the C1/2 lateral mass.

Case Report

An 83-year-old man with a 20-year history of RA complained of severe neck pain. Three months later, he complained of severe vertigo, with no visual field defect, when he leaned his head in the left-anterior direction. Because of his severe left occipital pain, he was able to sit for less than 10 minutes. When he lay on his left side, he always experienced vertigo.

There was no sign of a neurological defect, including cervical myelopathy or brain infarction, in the neutral position. Lateral radiographs indicated mild AAI: the atlantoaxial distances in the neutral, flexion, and extension positions were 4 mm, 5 mm, and 0 mm, respectively, and his Redlund–Johnell value in the neutral position was 27 mm, indicating mild AAI with mild vertebral subluxation (Fig. 1A–D). Enhanced cervical CT in the neutral

position indicated the destruction of the left lateral masses of C1/C2 (Figure 2A), and reconstructed three-dimensional CT angiography showed that the left VA was pinched between the posterior rim of the transverse foramen of C1 and the transverse process of C2, despite the dominance of left VA (Figure 2B–F). Doppler ultrasonography (Prosound α10, Aloka Co. Ltd, Tokyo, Japan) also visualized the positional occlusion of the left VA: the peak systolic velocities (PSVs) in the neutral, flexion, and traction positions were 23, 9, and 31 cm/s, respectively (Figure 3A–C), whereas the PSV of the right VA was almost 47 cm/s in any position, indicating that the left VA was occluded at the position that most readily induces vertigo.

We performed a posterior O–C2 fusion at the reduced position with an iliac bone graft (Figure 4A) instead of a C1–C2 fusion. It was difficult to achieve a secure grip on both C1 and C2 because of the severe destruction of the left lateral masses of C1 and C2. Therefore, a posterior transarticular screw was inserted on the right, and a C2 laminar screw and hook was used as an anchor because there was no space for a left transarticular screw or a C2 pedicle screw. The patient's severe vertigo disappeared in all positions immediately after the operation and there was no sign of postoperative infection or neurological defect. The patient was allowed to walk with a Philadelphia collar. The occlusion of the left VA disappeared completely (Figure 4B) and the PSV of the left VA recovered to 72 cm/s. Three months later, the O–C2 fusion was confirmed on CT and the patient reported no occipital pain or vertigo in any position.

Discussion

In previous reports of VA occlusion in patients with RA, angiography was used for the diagnosis.[3-12] Although angiography has many advantages, it is invasive and it does not

readily show the three-dimensional relationships between the arteries and the bone structure. In our patient, three-dimensional CT angiography in the neutral position, but not in the inducible position, demonstrated by chance a severe VA occlusion between the transverse foramens of C1 and C2. Doppler ultrasonography then revealed positional changes in the blood flow in the left VA and its complete occlusion when the patient leaned his head in the left-anterior direction. Three-dimensional CT angiography has been shown to have advantages in the detection of an abnormal course of the VA at the craniovertebral junction,[13,14] and is less invasive than angiography. However, it is difficult to obtain CT images of the arterial phase in various head positions. Color Doppler ultrasonography is noninvasive and can detect reduced blood flow in cervical spondylosis.[1] Furthermore, although ultrasonography is suitable for the detection of changes in the arterial flow that are dependent on the head position, it only poorly demonstrates the direct occlusion of the artery at C1/2. Therefore, the combination of ultrasonography and CT angiography is less invasive and more informative than angiography: ultrasonography used first to determine the head position that induces VA occlusion most, followed by CT angiography at the most inducible position, is strongly recommended.

The VA is divisible into four segments. The first segment runs from the VA origin to the transverse process. In the second segment, the VA ascends from the first transverse foramen (usually C6) to C3, then ascends laterally to the transverse foramen of C2. From here, the third segment emerges and sweeps laterally to pass through the transverse foramen of C1, and ends when the VA enters the dura. The rotational occlusion of the VA between C1 and C2 is known as “Bow Hunter’s stroke” and is considered physiological.[15] The usual explanation of the occlusion of the VA at C1–C2 during head turning is the stretching of the VA between the transverse foramens of C2 and C1.

Previous reports have attributed VA occlusion to osteophytic spurs or the stretching of the VA.[4] However, in our patient, three-dimensional CT angiography clearly showed that the distance between the left transverse processes of C1/C2 was severely reduced compared with that on the unaffected side (Fig. 2E and F), and that little osteophytic bony spur had formed about the transverse foramens (Fig. 2E). Because this space was reduced, the slight rotation and anterior shift of C1 easily led to the occlusion of the VA between the posterior rim of the transverse foramen of C1 and the transverse process of C2 (Fig. 2B and D). Generally, there is less bone formation in patients with RA than in those with other degenerative disorders. Therefore, in some previously reported cases of RA, the VA might have been pinched in a mechanism similar to that observed in our patient.

Because the one-sided collapse of C1 to C2 was the main factor underlying the occlusion of the VA, the causal treatment of this patient was to lift C1 against C2, rather than to remove the posterior rim of the transverse foramen of C1. The removal of bone is attended by the danger of VA injury, which must be avoided, especially on the dominant side, as in the present case. After the posterior fusion of O–C2 at the reduced position, the occlusion of the left VA disappeared completely.

In conclusion, we have described a patient with RA and positional occlusion of the VA, demonstrated by Doppler ultrasonography and CT angiography. Ultrasonography is suitable for the detection of positional VA occlusion and CT angiography for the description of the three-dimensional relationship between the occlusion and the bone structure. The one-sided destruction of the C1/2 lateral masses, associated with a slight anterior shift and rotation of C1, was considered to be the main mechanism of VA occlusion in this patient. This is the first report of such an application of this technology, and these methods may clarify the pathomechanism of this type of Bow Hunter's stroke when associated with RA.

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Figure legends

Figure 1

198 Lateral radiographs in the neutral (**A**), extension (**B**), and flexion (**C**) positions, and an
199 open-mouth view (**D**). Atlantoaxial instability and vertebral instability were mild. Note
200 that the left C1/2 joint space is not clear (**D**).
201
202 Figure 2
203 Coronal (**A**) and sagittal (**B**) CT angiographs, and a three-dimensional model of the arteries
204 only (**C**), the arteries with the bone structure (**D** and **F**), and the bones only (**E**). **A.** Note the
205 severe destruction of the left C1/2 lateral masses. **B** and **D**. The left VA is pinched between
206 the posterior rim of the transverse foramen of C1 (asterisk) and the transverse process of
207 C2. **C**. The arrowhead indicates the VA occlusion. **D–F**. Three-dimensional model of the
208 occipital and cervical bones (**E**) and showing the arteries (**D** and **F**). Asterisks indicate the
209 posterior rim of the transverse foramen of C1. Note that the left VA is dominant in **F** and
210 that the left distance between the C1 and C2 transverse processes is significantly shorter
211 than the right distance in **E** and **F**. Because of this reduced distance, a slight anterior shift
212 and rotation of C1 easily induces VA occlusion.
213
214 Figure 3
215 **A.** Doppler ultrasonography of the left VA at C3/4 with the blood velocity in the neutral
216 position. The peak of the wave indicates PSV. **B** and **C**. Blood velocities of the left VA with
217 flexion to the left-anterior direction (**B**) and in the traction position (**C**).
218
219 Figure 4
220 A lateral radiograph (**A**) and CT angiograph (**B**) after surgery. The arrowhead indicates the
221 decompressed left VA.

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224

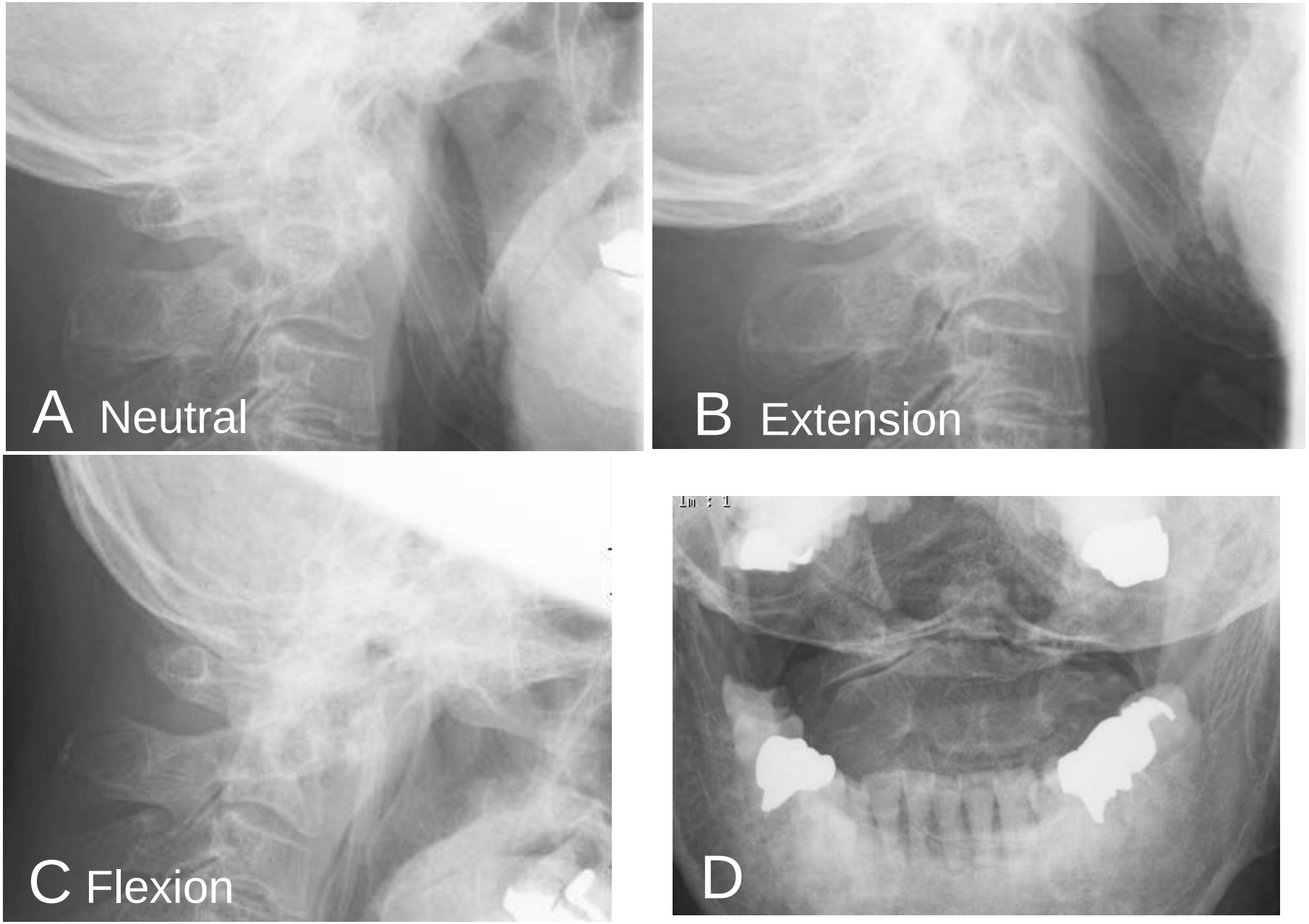

Figure 1

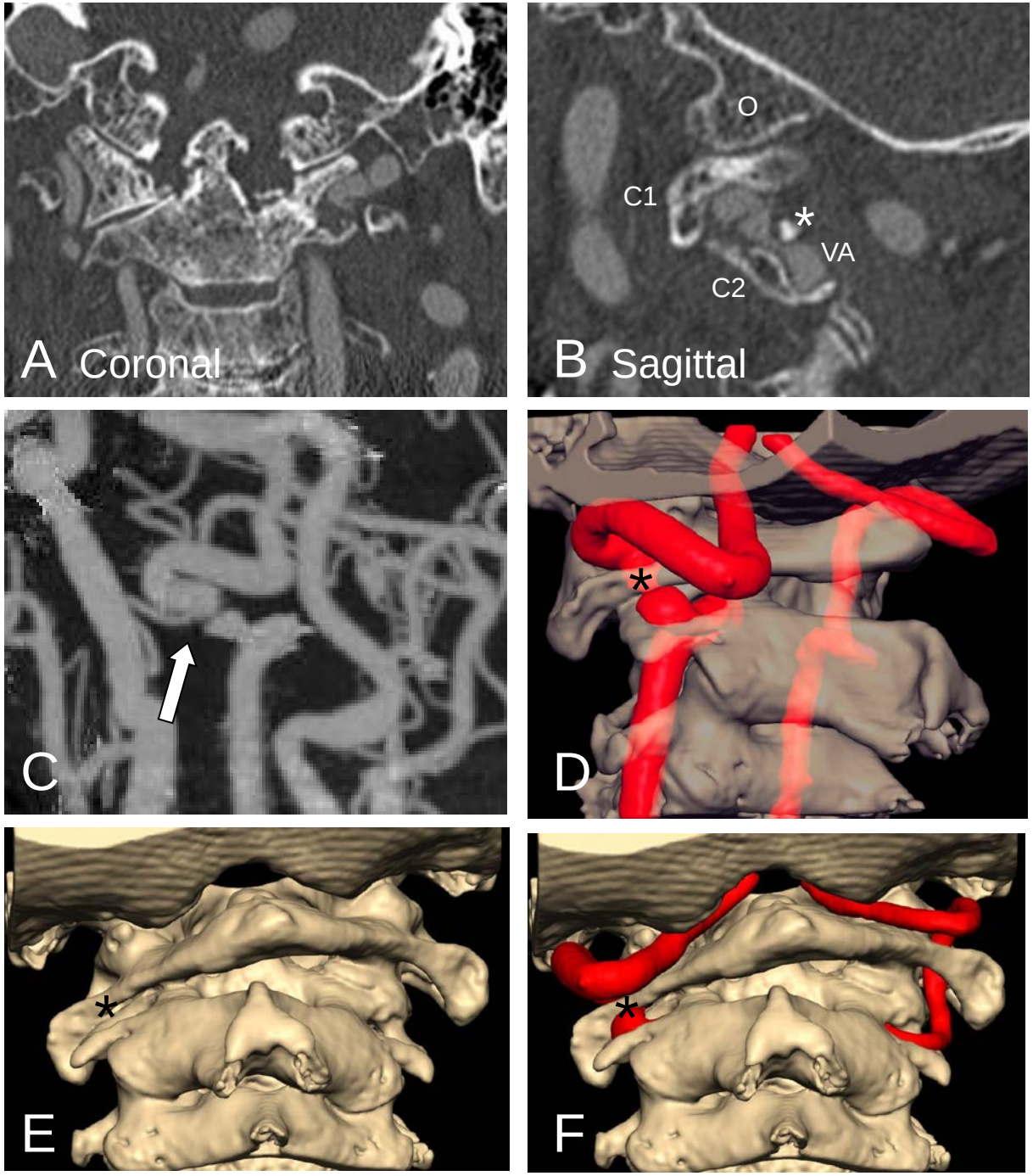

Figure 2

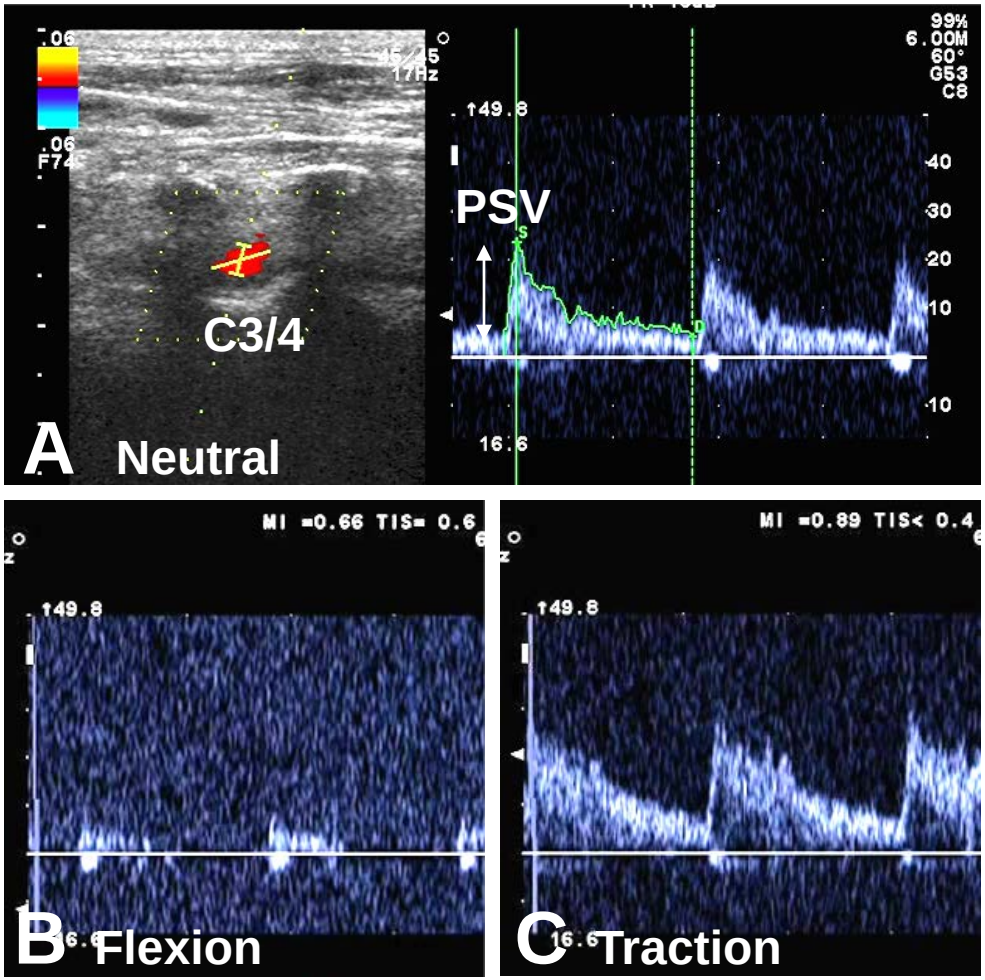
99%
6.00M
60°
G53
C8
149.8
PSV
40
30
20
10
-10
16.6
C3/4
A Neutral
MI =0.66 TIS= 0.6
149.8
B Flexion
MI =0.89 TIS< 0.4
149.8
16.6
C Traction

Figure 3

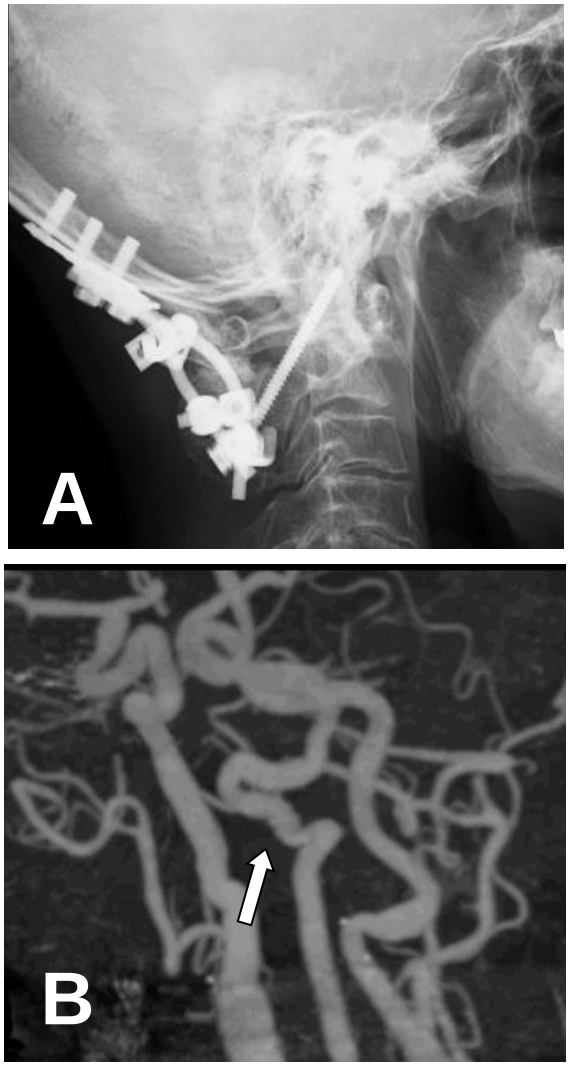

Figure 4