

Case Report

Two cases of multiple infarctions due to vertebral arterial occlusion associated with atlantoaxial subluxation caused by rheumatoid arthritis

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Case 1: A 58-year-old woman with rheumatoid arthritis (RA) suffered from pontine and cerebellar infarctions and underwent three-dimensional computed tomography angiography (3D-CTA) of the vertebral artery (VA) with left-sided rotation of the head. This revealed a destroyed right atlantoaxial (AA) joint and right VA occlusion, which had caused the infarction. No recurrent infarctions have been observed after transarticular screw fixation (Magerl technique) was performed. Case 2: A 68-year-old woman with RA also suffered from pontine and cerebellar infarctions and underwent 3D-CTA of the VA with right-sided rotation of the head. This revealed a destroyed left AA joint and left VA occlusion, which had caused the infarction. No recurrent infarctions have been observed after posterior fusion between the occiput and 3rd cervical vertebra was performed.

In both cases of RA, who had multiple infarctions due to VA occlusion and a temporary thrombus formation, 3D-CTA in a rotated position helped to diagnose arterial occlusion. In addition, posterior fusion of the AA joint was a useful surgical technique.

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Introduction

Rheumatoid arthritis (RA) is associated with a high incidence of atlantoaxial subluxation (AAS)¹. Because the vertebral arteries (VA) are near the atlantoaxial (AA) joint, VA occlusion may occur due to AAS. Since the VA supplies blood to the cerebellum and brainstem, serious symptoms can develop if infarction occurs due to VA occlusion. These types of lesions have previously been reported^{2,3}. Because of the difficulty of making the diagnosis with computed tomography (CT) and magnetic resonance angiography (MRA), a

definitive diagnosis cannot be made in many cases. In this report, two cases of multiple infarctions due to VA occlusion and a temporary thrombus formation associated with AAS are described. In these patients, 3D-CTA of the VA during rotation was useful for making a definite diagnosis.

Case reports

Case 1

A 58-year-old woman was diagnosed with RA 8 years ear-

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lier. She experienced neck pain and was diagnosed with AAS 2 years earlier. Six months later, she experienced dizziness and nausea during neck flexion, and she then visited our hospital. Detailed examinations revealed pontine and cerebellar infarctions. On physical examination, other than increased tendon reflexes in all four extremities, there were no other abnormalities.

Cranial magnetic resonance imaging (MRI) with T2*-weighted imaging and diffusion-weighted imaging revealed fresh infarcts in the pons and cerebellum (Fig. 1, a, b). On the lateral view X-ray of the cervical spine, there was no abnormal motion. On CT of the cervical spine, there was destruction of the AA joint, predominantly on the right (Fig. 1, c). Cervical MRI revealed mild stenosis of the spinal canal at the level of the atlas and C6/7 level (Fig. 1, d). When 3D-CTA of the VA was performed, there was slight right rotation within the AA joint, but no occlusion of the VA was observed (Fig. 2, a). 3D-CTA with maximum left rotation of the head showed right VA occlusion due to destruction of the right AA joint (Fig. 2, b, c, d).

The patient was diagnosed as having infarction due to VA occlusion with rotation of the AA joint. Transarticular screw fixation (Magerl technique)⁴⁾ with a hydroxyapatite insert was performed (Fig. 3,a,b). Currently, 2.5 years after surgery, her clinical course has been good with no recurrent infarctions.

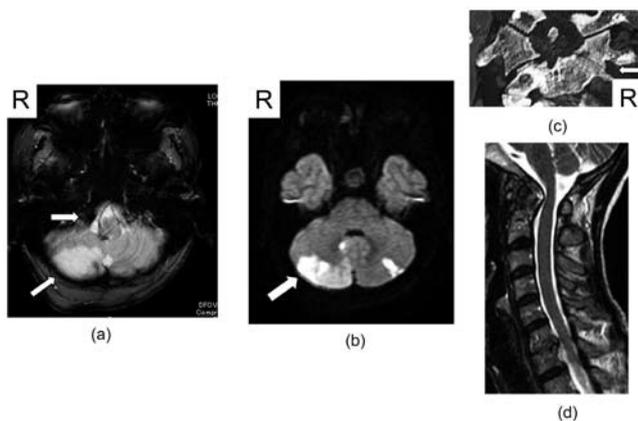


Fig. 1 (Case 1)

A 58-year-old woman

(a, b) On cranial MRI, T2*-weighted imaging and diffusion-weighted imaging reveals fresh pontine and right cerebellar infarctions (arrows). (c) CT of the cervical spine (Coronal), there is destruction of the AA joint, predominantly on the right (arrows). (d) MRI (T2 weighted images, Sagittal) shows mild spinal canal stenosis at the atlas and C6/7 level.

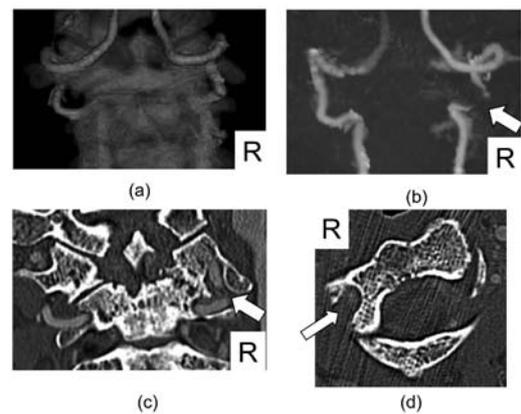


Fig. 2

(a) On 3D-CTA, there is slight right rotation within the AA joint but no occlusion of the VA is observed. (b) With maximum left rotation of the head, 3D-CTA shows right VA occlusion due to destruction of the right AA joint (arrows). (c) Coronal, (d) Axial (C1/2 level)

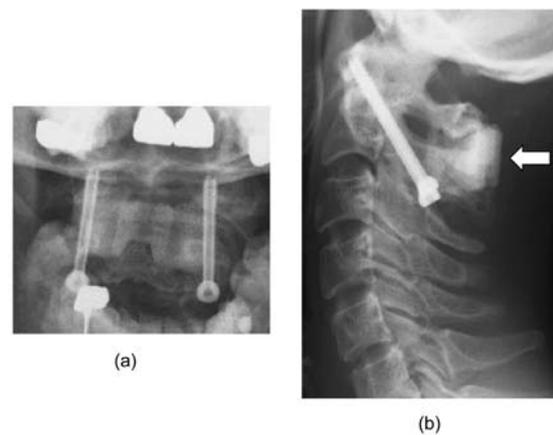


Fig. 3

Post surgical radiography. (a) Open mouth view. (b) Lateral view. Transarticular screw fixation (Magerl technique) was performed. The C1/2 spinous process is fixed with a hydroxyapatite insert (arrows).

Case 2

A 68-year-old woman was diagnosed with RA 23 years ago. Because of dizziness while sleeping at night, which worsened the next day, she was examined at the hospital where her RA was being controlled. Detailed examination revealed pontine and cerebellar infarctions. Since these symptoms improved gradually, the patient was left untreated. However, 2 months later, she again experienced dizziness with new infarctions of the pons and cerebellum. The patient visited our hospital. On physical examination, neck tenderness and abnormal reflexes of all four extremities were observed, but no other abnormalities were noted.

Cranial MRI with T2-weighted imaging revealed fresh pontine and cerebellar infarctions (Fig. 4, a). Twelve day after, there were new fresh left cerebellar infarctions (Fig. 4, b). On cervical radiography and CT, destruction of the AA joint, predominantly on the left, was observed (Fig. 4, c). Cervical MRI showed no stenosis of the spinal canal (Fig. 4, d). On 3D-CTA, there was left rotation within the AA joint with slight deviation, but no occlusion of the VA was observed (Fig. 5, a). Therefore, to further evaluate the infarctions, when 3D-CTA was performed with maximum right rotation of the head, left VA occlusion due to left AA joint destruction was observed (Fig. 5, b, c, d).

As in case 1, this patient was also diagnosed as having infarction due to VA occlusion with rotation of the AA joint. Posterior fusion between the occiput and 3rd cervical vertebra was performed (Fig. 6,a). Postoperatively, on 3D-CTA with maximum right rotation, no VA occlusion was noted (Fig. 6,b). One year after surgery, her clinical course has been good with no further recurrent infarctions.

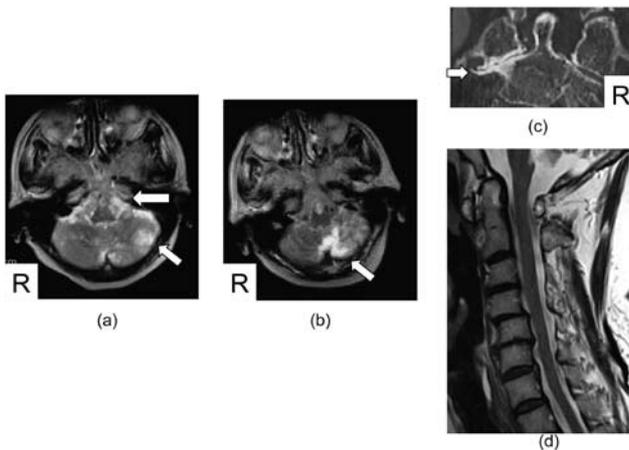


Fig. 4 (Case 2)
 A 68-year-old woman
 (a) On cranial MRI, T2-weighted imaging, there are fresh pontine and left cerebellar infarctions (arrows). (b) Twelve day after, there are new fresh left cerebellar infarctions (arrows). (c) CT of the cervical spine, there is destruction of the AA joint, predominantly on the left (arrows). (d) MRI shows no stenosis of the spinal canal.

Discussion

In 1968, Webb et al.³⁾ reported an autopsy case in a patient who died due to VA insufficiency associated with a cervical spine lesion due to RA. In addition, in 1978, Sorensen²⁾ reported a case of bow hunter’s stroke in a patient with a brain

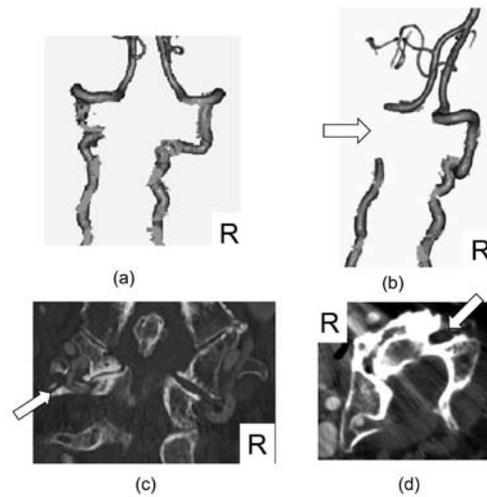


Fig. 5
 (a) On 3D-CTA, there is left rotation within the AA joint with slight deviation, but no occlusion of the VA is observed. (b) On 3D-CTA, with maximum right rotation of the head, there is left VA occlusion due to destruction of the left AA joint (arrows). (c) Coronal, (d) Axial (C1/2 level)

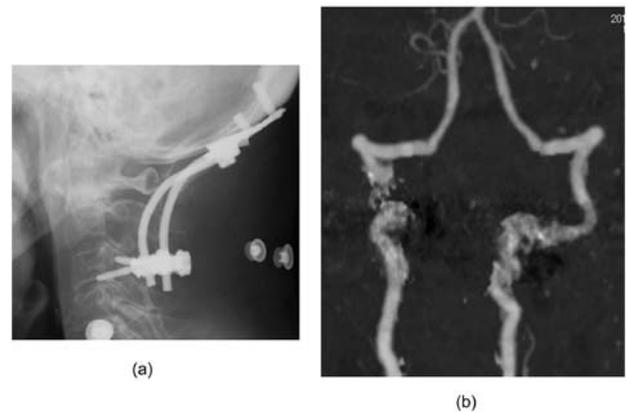


Fig. 6
 Post surgical radiography. (a) Lateral view. Posterior fusion between the occiput and 3rd cervical vertebra has been performed. (b) Postoperatively, on 3D-CTA with maximum right rotation, no VA occlusion is observed.

stem infarction while shooting arrows during archery practice. Gaikwad et al.⁵⁾ reported an RA patient who had multiple infarctions. Although there are a few other similar reports^{6,7)}, this is a relatively rare disorder. In the present report, two cases of multiple infarctions with a temporary thrombus formation due to AAS associated with RA were described.

Cerebral angiography is useful in the diagnosis of VA occlusion^{5,8)}. However, this examination is highly invasive, burdensome for patients, and does not depict relationships with bony structures. From this standpoint, 3D-CTA with

head rotation is highly useful for making a definitive diagnosis of VA occlusion. 3D-CTA can be performed as an outpatient, and the site and cause of VA occlusion can be diagnosed accurately. In both of the present patients, VA occlusion lateral to the AA joint could be diagnosed. As in the present patients, when symptoms develop due to infarction, detailed examination can establish a diagnosis, but in patients who have transient symptoms or are asymptomatic, the diagnosis is likely to be difficult. Patients with cervical spine lesions due to RA should be carefully asked about transient symptoms such as dizziness. If VA occlusion is suspected, 3D-CTA in a rotated position should be performed to diagnose VA occlusion.

Treatment for VA occlusion due to AAS includes posterior fusion of the AA joint^{7,9)} and decompression of the cause of VA occlusion¹⁰⁻¹³⁾. Because decompression does not stabilize the AA joint, rotational mobility can be maintained postoperatively. However, Matsuyama et al.¹⁴⁾, in a comparison between posterior fusion and decompression, reported restenosis after decompression. They also reported better patient satisfaction with posterior fusion than with decompression. In the present report, the patients also had a good clinical course without recurrent infarctions after posterior fusion. We believe that posterior fusion of the AA joint is a useful surgical technique.

Conclusion

Two patients with RA who had multiple infarctions due to VA occlusion and a temporary thrombus formation were reported. In these cases, 3D-CTA in a rotated position was helpful for making a definitive diagnosis. Posterior fusion of the AA joint is a useful surgical treatment.

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