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## Case Report

# Vertebral and carotid artery aneurysms and polyarthritis in a patient with Takayasu arteritis: A case report ☆☆☆☆

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## ABSTRACT

Takayasu arteritis (TA) is a common autoimmune disease in the clinical setting. However, vertebral artery aneurysms caused by TA are rarely reported. We herein describe a 28-year-old man with multiple vertebral artery aneurysms and carotid artery aneurysms caused by TA, which showed typical wall thickening and lumen dilation with a “string of beads” appearance by Doppler ultrasound and radiology. Previous studies have shown that most TA-associated vertebral artery lesions are stenosis, occlusion, and dissection of the intracranial part of the artery. In this case, TA mainly affected the cervical segment of the vertebral artery (the intracranial segment was not obviously involved), and the main manifestations were aneurysms and occlusion. This case provides more information for further understanding of TA-associated vertebral artery lesions.

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## Introduction

Takayasu arteritis (TA) is a type of chronic nonspecific macrovascular inflammation characterized by arterial segmental stenosis, occlusion, dilation, or aneurysm formation

[1]. TA mainly occurs in women of childbearing age. The male:female ratio is 1.0:6.6. TA can be divided into 4 types [2]: type I, brachiocephalic artery type; type II, thoracoabdominal aorta type; type III, extensive type; and type IV, pulmonary artery type. Vertebral artery aneurysms caused by TA are rarely reported. We herein present a clinical case of multiple vertebral

☆ The first two authors contributed equally to this work and should be considered co-first authors.

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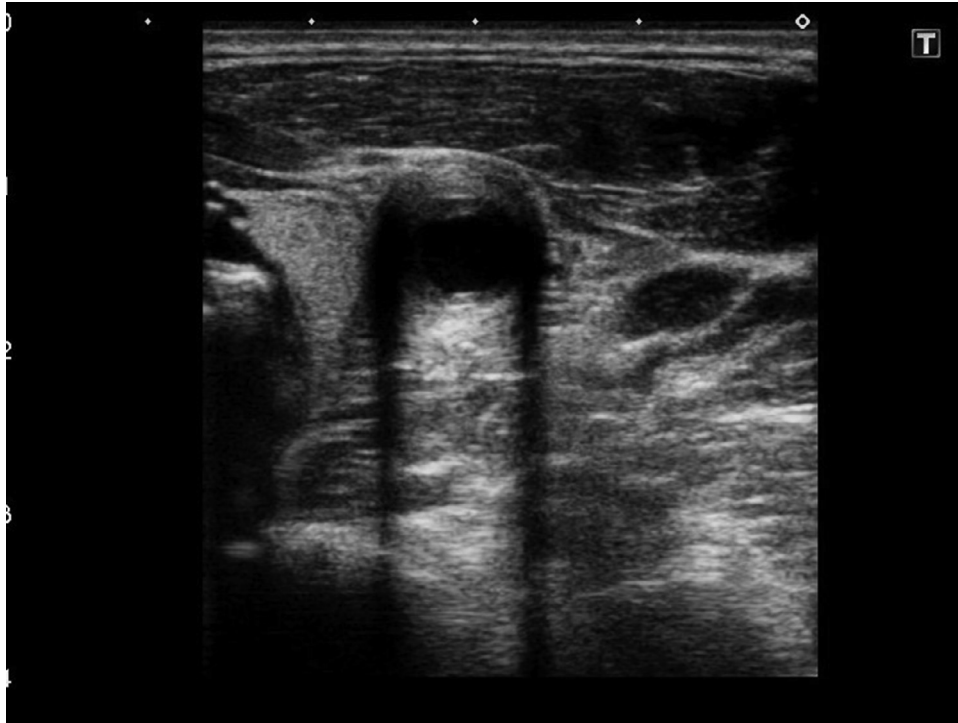
\* Patient consent: Written informed consent for publication was obtained from the patient.

\* Corresponding author

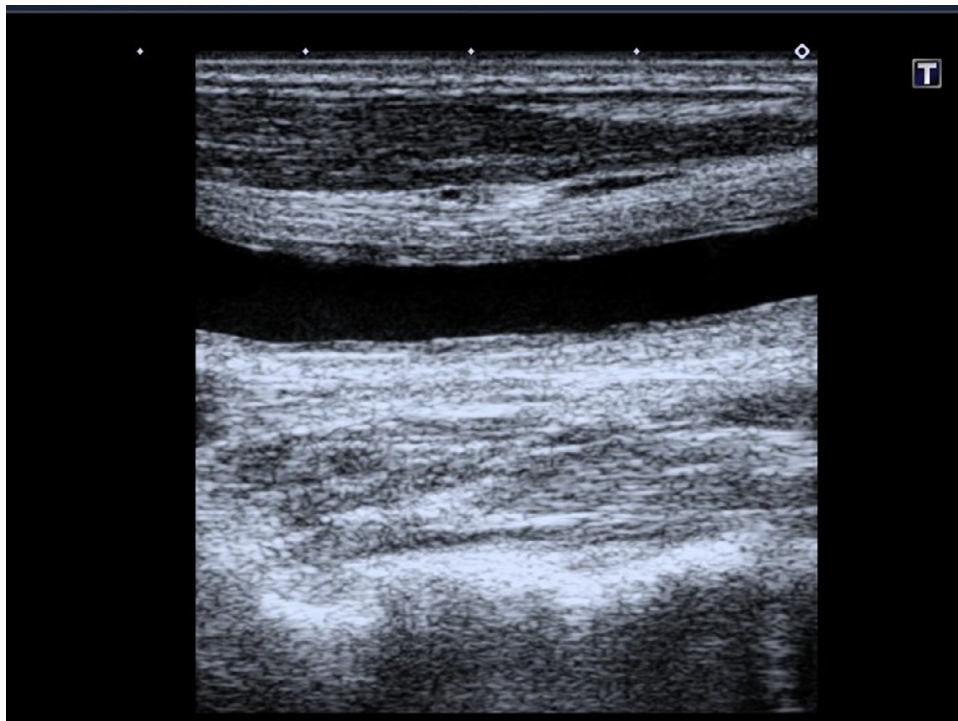
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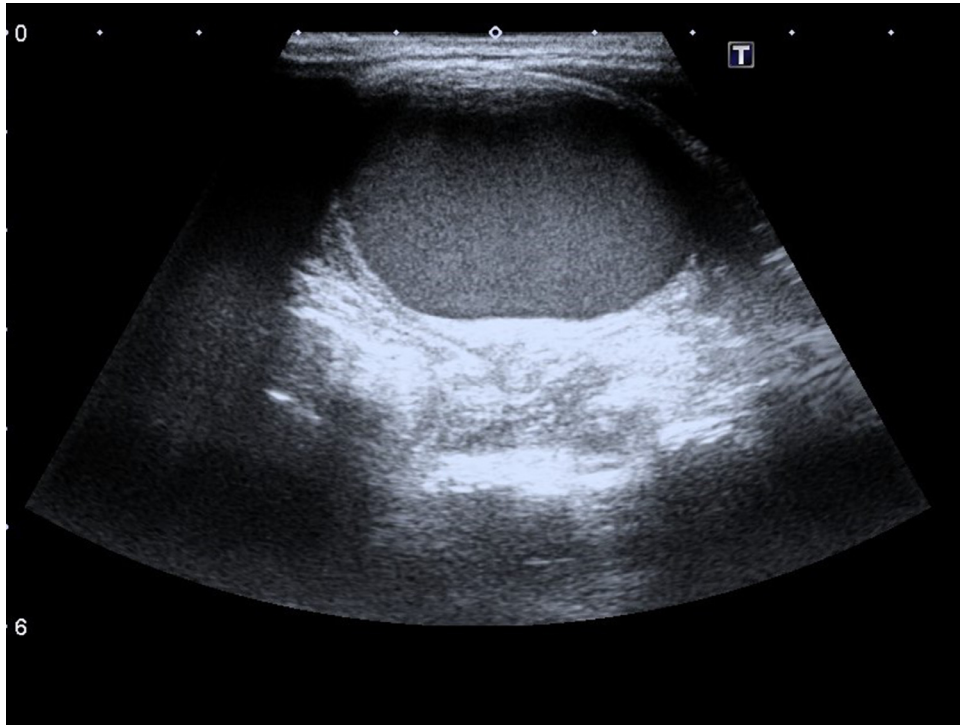
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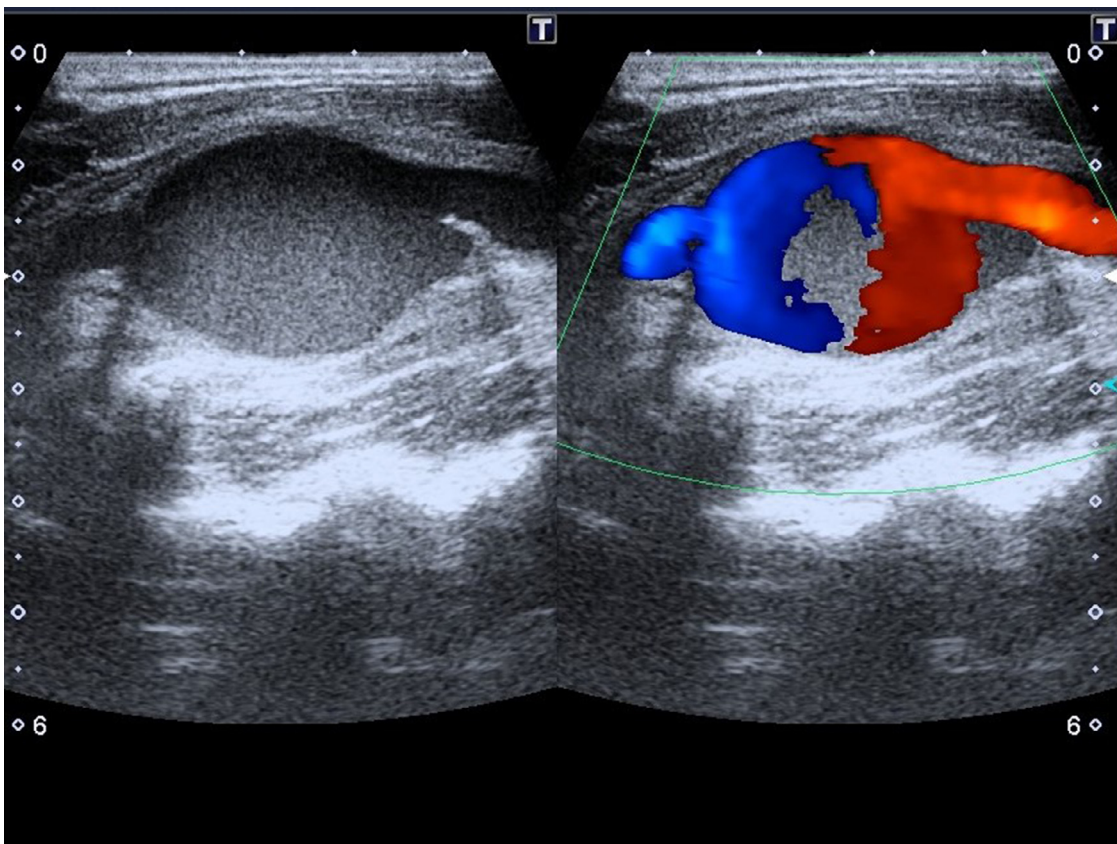
**Fig. 1** – Cross section of carotid artery. The wall of the carotid artery was thickened, and the anterior wall showed “onion skin” -like changes.



**Fig. 2** – Longitudinal section of the common carotid artery. The wall was thickened, and scattered punctate hyperechoic calcifications were present in the intimal layer.



**Fig. 3** - Aneurysms at the left common carotid artery bifurcation. The walls of the aneurysms were thin, and the blood flow in the aneurysms showed cloud-like spontaneous development.



**Fig. 4** - The left vertebral artery lumen was dilated and showed an aneurysm of about 2.9 x 1.8 cm, and the internal blood flow exhibited whirling eddy-like movement.



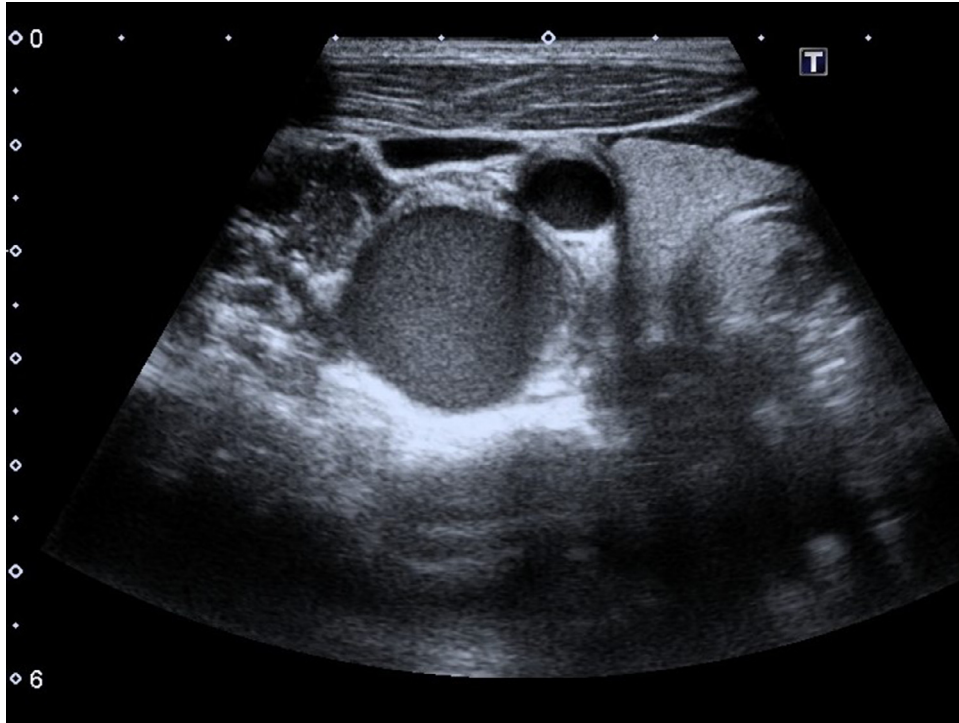


Fig. 5 – The anteromedial part of the aneurysm was present in the right common carotid artery.

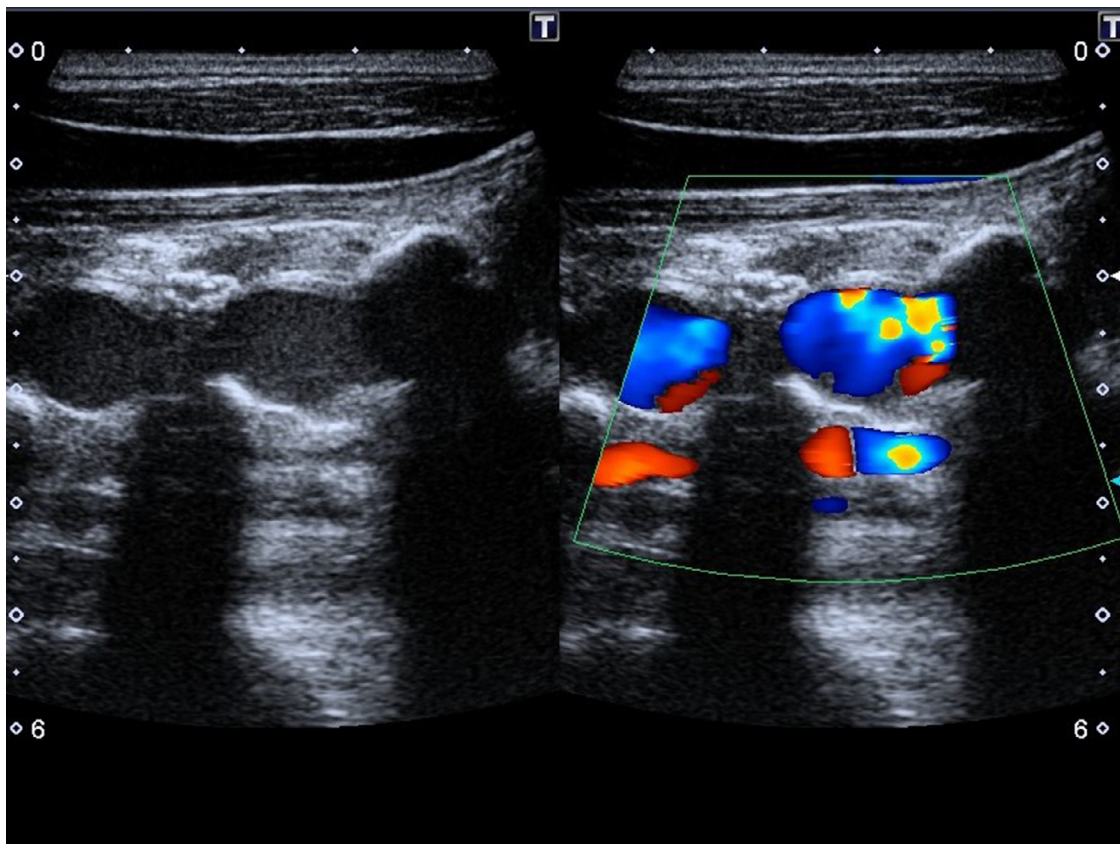
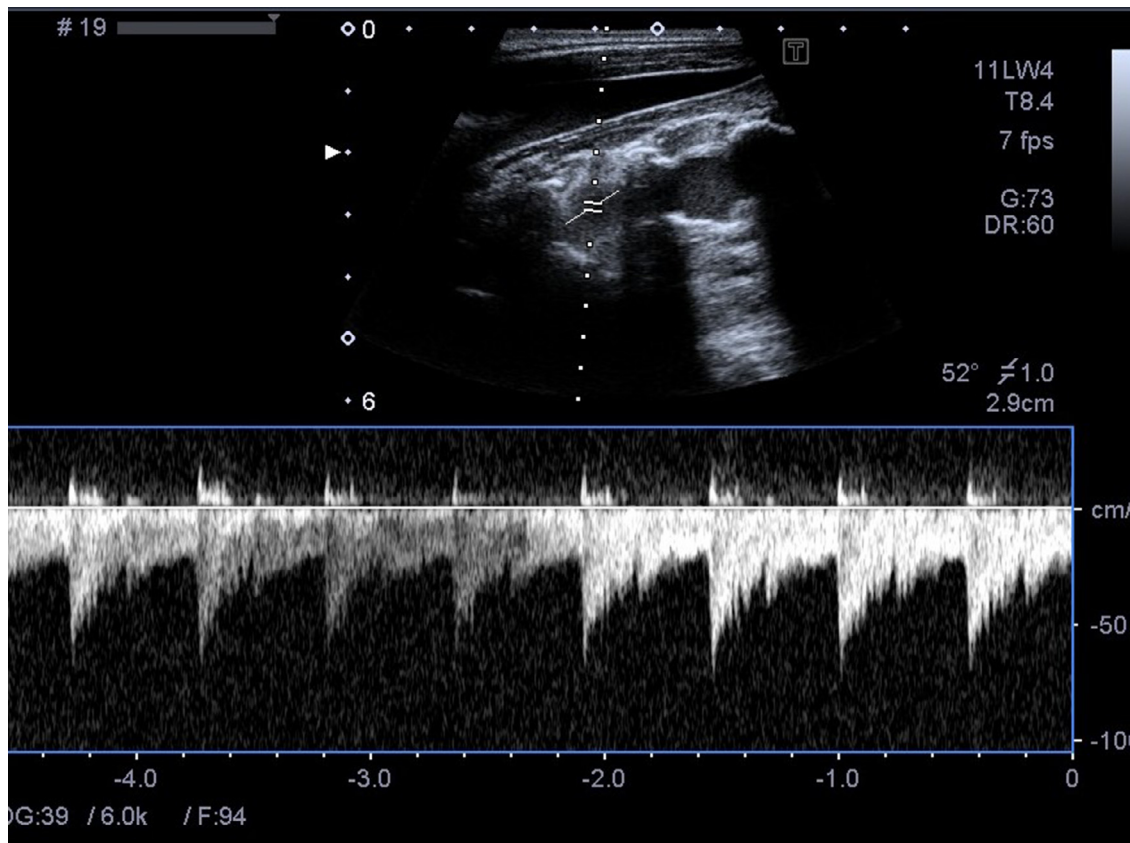


Fig. 6 – The lumen of the intervertebral segment of the left vertebral artery showed frequent dilations, resulting in a “string of beads” appearance. On color Doppler flow imaging, the internal blood flow signal was colorful, and had lost its laminar flow characteristics.



**Fig. 7 – Doppler spectrum of blood flow in the intervertebral segment of the left vertebral artery. The frequency window had disappeared and the frequency band showed a widened, hairy, and spiny appearance.**

artery aneurysms and carotid artery aneurysms caused by TA and discuss the pathology and medical imaging characteristics combined with a brief review of the literature.

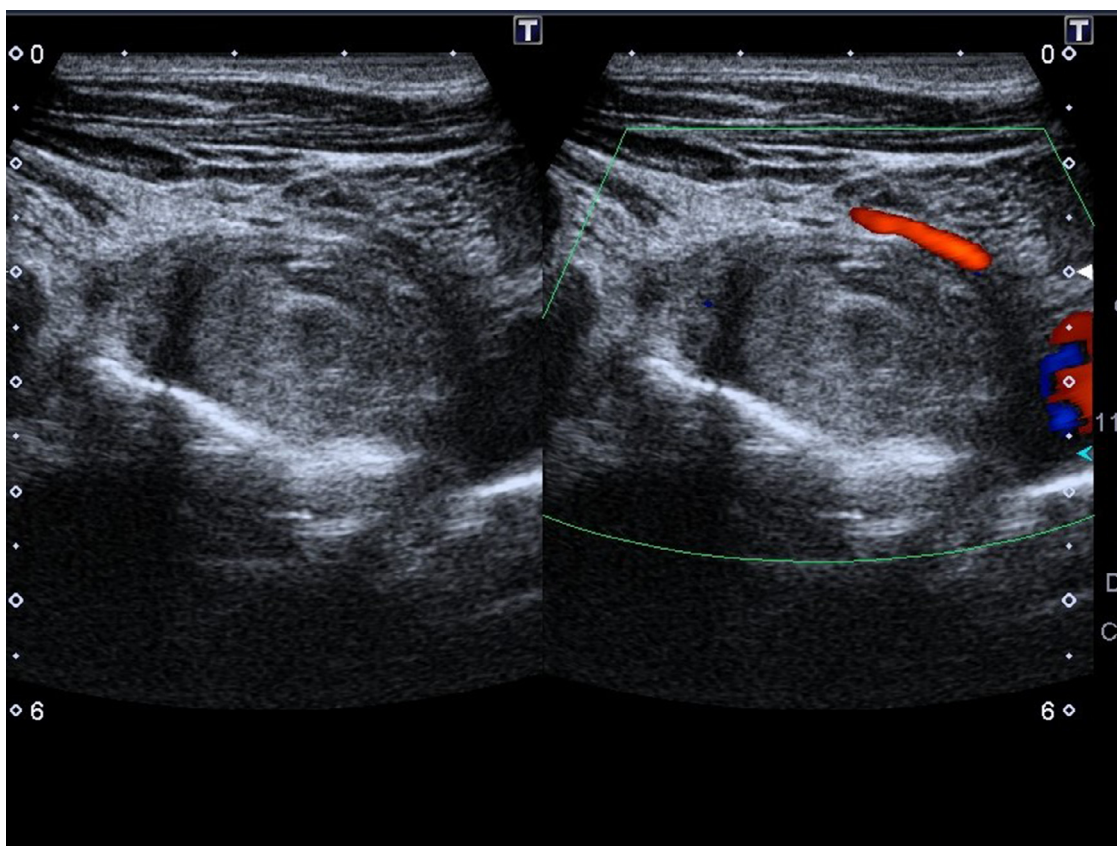
### Case description

A 28-year-old man presented to our hospital with a 6-month history of intermittent multiple joint swelling and pain that had become aggravated during the past week. He was in good health and denied a history of infectious or chronic disease. Physical examination revealed that the patient had mild polyarticular swelling and limited movement of the bilateral ankle and knee joints, and he reported morning stiffness for about 10 minutes that was relieved after exercise. Schober's test was positive (2.5 cm). The left cervical lymph node was enlarged at  $3.5 \times 4.5$  cm, and a murmur was heard in the left neck. The patient's body weight had decreased by 7.5 kg during the previous 2 months.

Ultrasound examination of the neck blood vessels revealed 4 major findings. First, the intima-media layer of the bilateral carotid arteries was unclear, the vascular wall was thickened (Figs. 1, 2) (the thickest part was 2.7–3.2 mm), the intima

was not smooth, and multiple punctate hyperechoic lesions were present. Second, the lumen of the bifurcation of the left common carotid artery and the initial segment of the external carotid artery was dilated like a tumor, with a range of about  $3.6 \times 1.8$  cm (Fig. 3). The blood flow inside the tumor was turbulent. Third, multiple aneurysms were present in the right cervical segment of the vertebral artery (the largest was about  $2.9 \times 1.8$  cm), and the blood flow in the aneurysms was colorful and disordered (Figs. 4–7). Fourth, the carotid segment of the left vertebral artery was dilated to about  $3.3 \times 1.9$  cm, consistent with an aneurysm. The tumor was full of low and medium echo and had no blood flow signal. The lumen of the vertebral segment was also dilated locally to the level of the carotid bifurcation, and no blood flow was present in the left vertebral artery lumen (Fig. 8).

CT angiography (CTA) of the carotid artery (Figs. 9, 10) showed multiple aneurysms at the bifurcation of the left common carotid artery, the proximal part of the left external carotid artery, the proximal part of the right vertebral artery, and the cervical spine. The lumen of the proximal part of the left vertebral artery was enlarged, and the distal part of the vertebral artery was not visualized. A 3-dimensional reconstruction map of CTA (Figs. 11, 12) showed multiple aneurysmal dilations in the right cervical segment of the vertebral



**Fig. 8 – The left vertebral artery was locally dilated in the shape of a tumor, measuring about 3.3 x 1.9 cm. The tumor-like lesion was moderately filled. Color Doppler flow imaging showed no blood flow signal, and a thrombus was present.**

artery arranged in a “string of beads” pattern, with the largest located at the root of the neck and aneurysmal dilations present at the bifurcation of the left carotid artery.

Laboratory examination revealed the following: microcytic anemia with a hemoglobin concentration of 8.4 g/dL; erythrocyte sedimentation rate, 93 mm/h; C-reactive protein, 75.80 mg/L; immunoglobulin G, 15.70 g/L; HLA-B27, negative; rheumatoid factor, negative; and tuberculosis diagnostic test, negative. A full-body bone scan showed multiple arthritis of both knees and left ankles (Fig. 13).

According to the diagnostic criteria for TA established by the American Rheumatic Association in 1990, this patient’s clinical manifestations, laboratory examination findings, and imaging data led to a diagnosis of (i) TA complicated with vertebral aneurysm and carotid aneurysm formation and (ii) TA-associated arthritis.

## Discussion

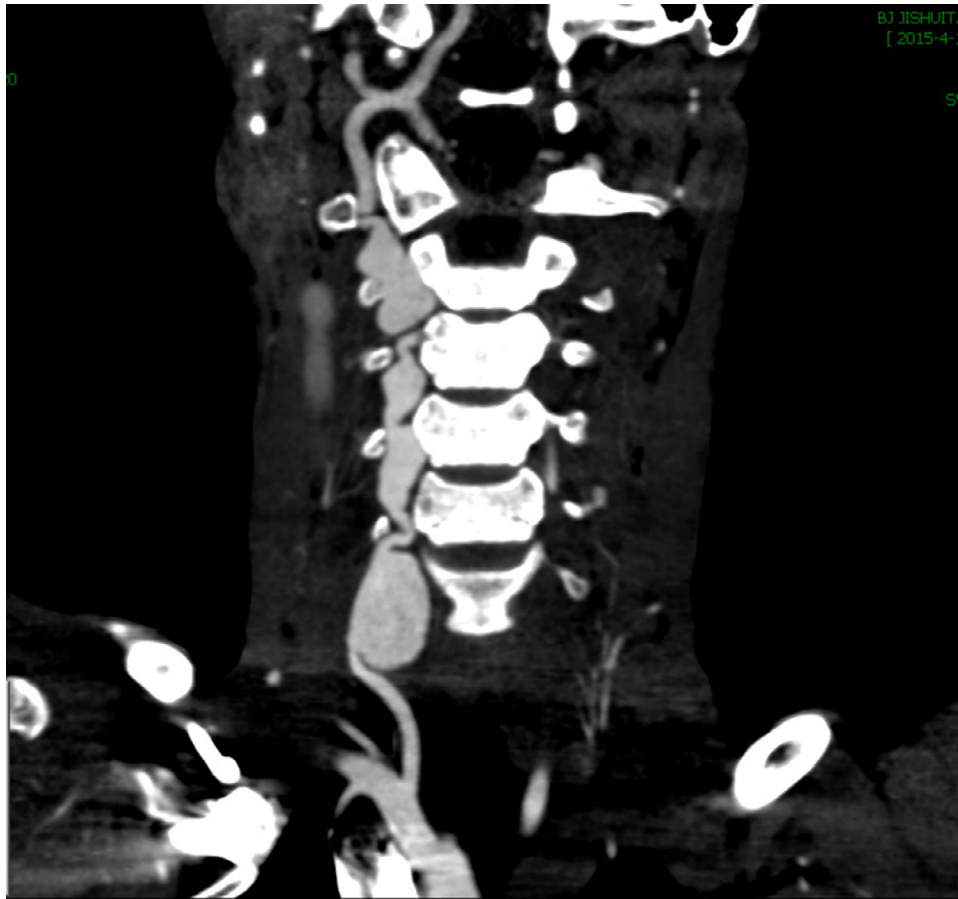
TA mostly involves the aorta and its primary branches. TA reportedly involves the subclavian artery in about 65.0%-79.8% of cases, the carotid artery in 43.0%-79.1%, and the vertebral artery in 19%. Intracranial stenosis and dissection are common, whereas cervical vertebral artery aneurysms are rare

[3]. Vertebral aneurysms caused by TA are rarely reported, and multiple vertebral aneurysms in the cervical segment have not been reported to date.

TA is full-thickness arteritis involving intimal damage [4], a broken or absent elastic plate, and sometimes a large amount of mucopolysaccharide accumulation, forming a “mucus lake” shape. In old lesions, new capillaries appear in the thickened intima, and calcification is seen in some cases [5]. The elastic fibers and smooth muscle tissue of the involved artery become damaged and necrotic and even form focal fibrous scars; these changes are accompanied by the formation of focal inflammatory granulomas as well as scattered inflammatory lesions infiltrated by lymphocytes, monocytes, and multinucleated macrophages. Dense connective tissue hyperplasia is often present in the adventitia, resulting in obvious adventitial thickening. In addition, inflammatory lesions in the intima and media lead to thickening of the vascular wall and stenosis or even occlusion of the lumen. The cross section shows “onion-like” layer-by-layer thickening of the vascular wall [6]. These pathologic changes lead to impaired elasticity of the wall, resulting in stenosis, dilatation, occlusion, rupture, and aneurysm formation.

The above pathologic changes may have corresponding imaging findings, such as the “onion skin”-like changes in ultrasound cross section; calcification of old lesions; multiple punctate hyperechoic lesions in ultrasound imaging; and





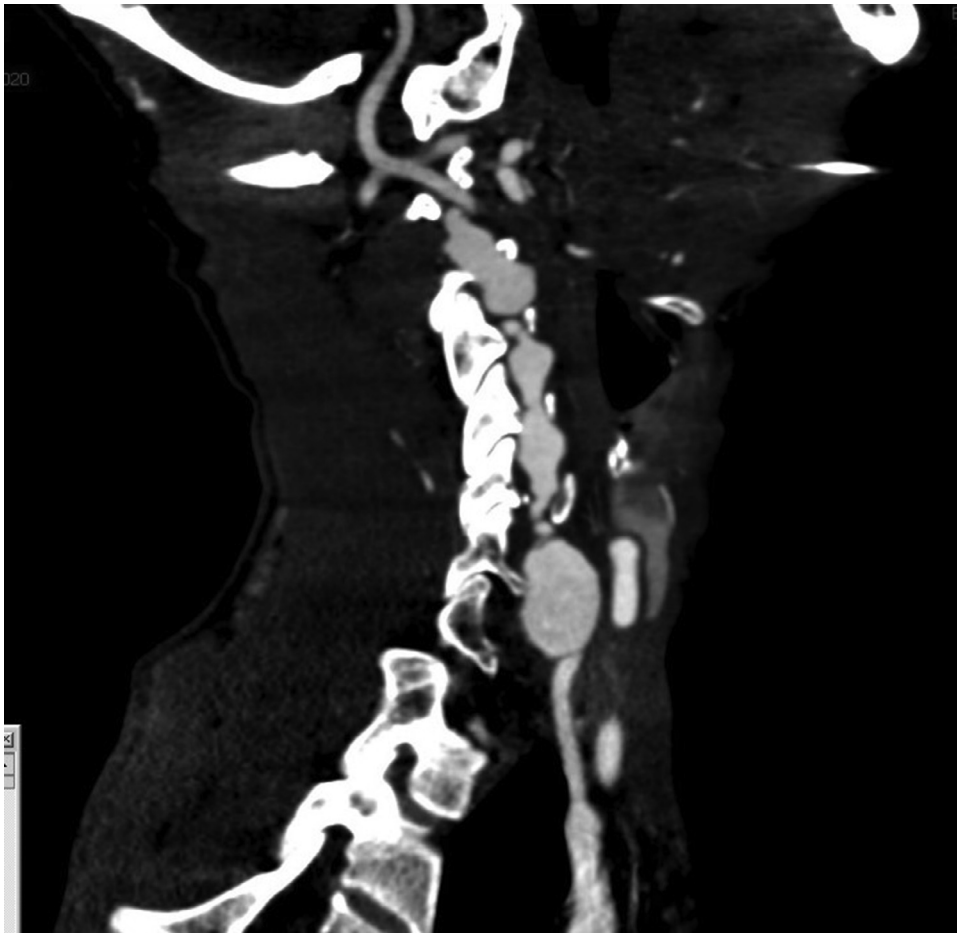
**Fig. 9 – Computed tomography angiography showed multiple aneurysms in the right vertebral neck and vertebral segment.**

stenosis and expansion of the lumen. These changes can be detected by spectral Doppler, which displays the blood flow, velocity, and acceleration time in the lumen; such examination is helpful to evaluate the hemodynamics of the involved vessels. Ultrasonic examination can show the distribution of neovascularization in the lumen and wall of the involved vessels [7]. In CTA imaging, the walls of TA-affected arteries are thickened to varying degrees with mild enhancement or no enhancement. The lumen may also be occluded or stenosed to varying degrees. The lumen stenosis is centripetal, and some vessels become dilated after stenosis. In patients with active TA, the arterial wall can show layered thickening; the outer layer shows high density, and the inner layer shows low density with mild enhancement, which is closely related to the above-described pathologic changes [8]. Magnetic resonance imaging with CTA 3-dimensional reconstruction technology is a volume scanning method, which has the advantages of a wide display range, high image resolution, and fast scanning speed. It can show lesions in 3 dimensions and multiple directions and can visually display the lesion location, involved range, wall changes, collateral circulation, and lumen changes in patients with TA. The axial image facilitates comprehensive evaluation of the thickening state of the lesion wall. Conventional digital subtraction angiography is the clinical gold standard diagnostic technique for TA because it can accurately reflect the dynamic characteristics of the vas-

cular morphology. It has the clinical advantages of high resolution of small vessels and clear images. Contrast filling images provide high-density resolution and spatial resolution [9]. However, this technique can only show the location, scope, and degree of stenosis; it cannot show the specific pathologic changes of vascular wall thickening.

In this case, the bilateral carotid arteries were extensively involved, left common carotid artery and external carotid artery aneurysms were present, the bilateral vertebral arteries showed multiple aneurysms with a “string of beads” appearance, some aneurysms were combined with thrombosis, the ultrasound images showed “beaded” expansion, and the internal blood flow was turbulent and cloud-like on imaging; these are typical manifestations of aneurysms. At the first visit, the left and right vertebral artery aneurysms were  $3.3 \times 1.9$  and  $2.9 \times 1.8$  cm, respectively. At 12 days and 30 days after treatment for TA, both of the vertebral artery aneurysms had become smaller. The ultrasound examination showed carotid artery and vertebral artery wall thickening as well as multiple aneurysm formation, providing the most intuitive evidence for the diagnosis of TA.

In conclusion, color Doppler ultrasound is a noninvasive examination method with high resolution that can quickly evaluate the wall, lumen, and blood flow of TA-affected vessels [10]. It can also repeatedly and dynamically detect the progress of pathologic changes.

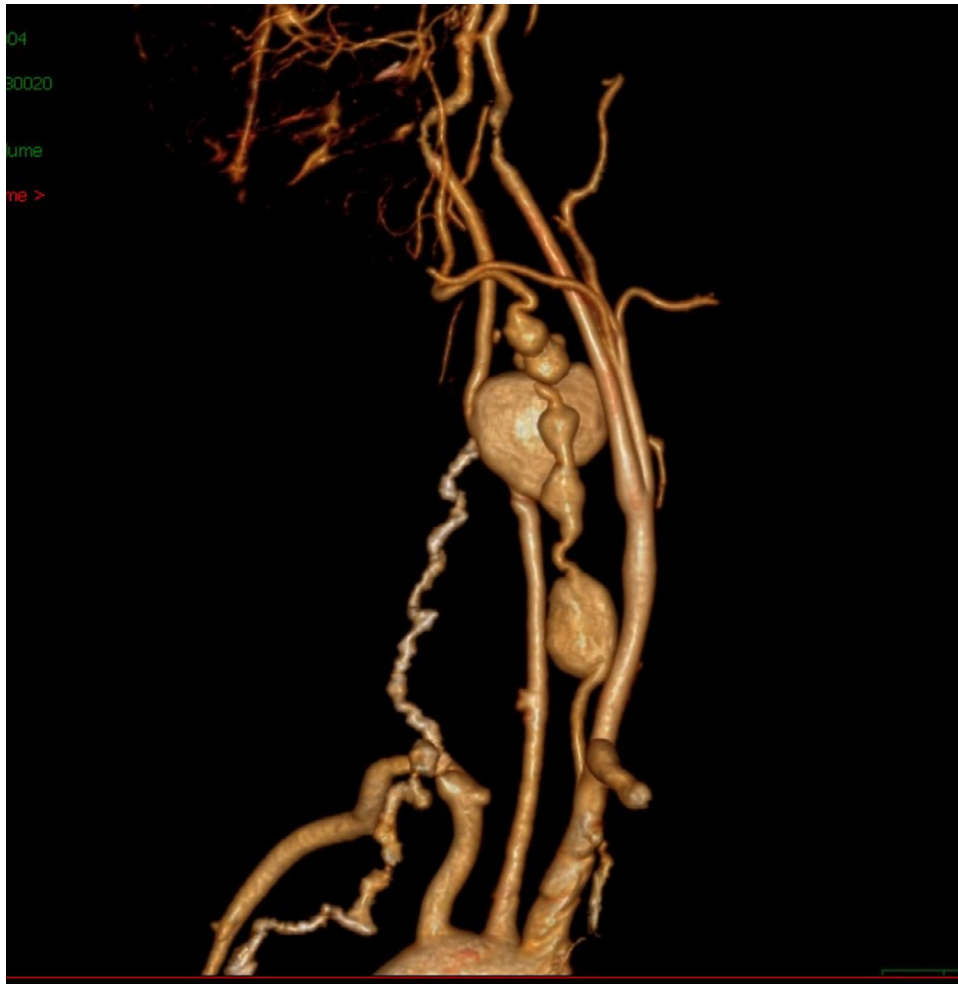


**Fig. 10 – Sagittal view of computed tomography angiography showed multiple aneurysms in the right vertebral artery neck and vertebral segment.**

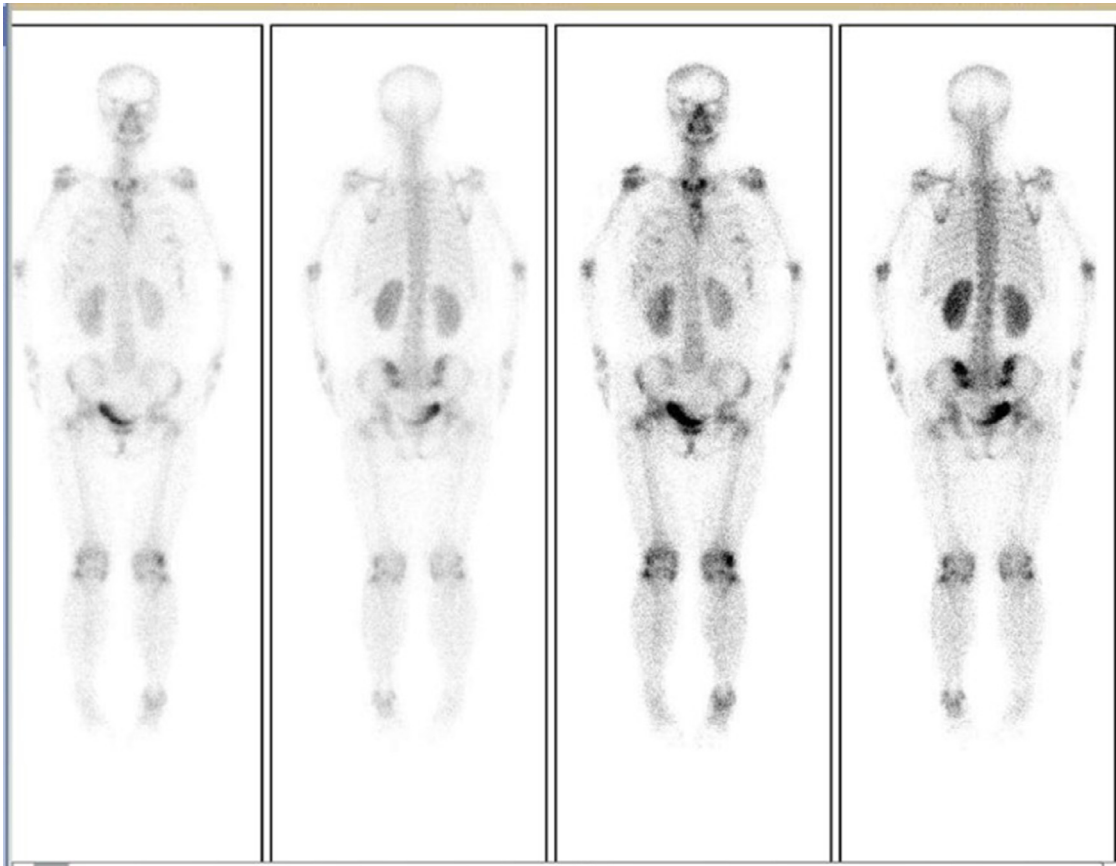




**Fig. 11** – Computed tomography angiography positive bitmap showed multiple aneurysmal dilations in the right cervical segment of the vertebral artery arranged in a “beaded” pattern, with the largest located at the root of the neck; additionally, aneurysmal dilations were present at the bifurcation of the left carotid artery. The left vertebral artery was dilated and the distal segment was undeveloped.



**Fig. 12** – Computed tomography angiography side map showed multiple aneurysmal dilations in the right vertebral artery carotid lumen arranged in a “beaded” pattern, the largest located at the root of the neck; additionally, the left carotid artery bifurcation showed aneurysmal dilation. The left vertebral artery was dilatated and the distal segment was undeveloped.



**Fig. 13 – A full-body bone scan showed multiple arthritis of both knees and left ankles. The radioactivity in bilateral knee joint, left ankle joint and other joints is unevenly increased, which is considered as arthritis.**

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