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Subclavian steal syndrome without Stenosis: A Case Report and Review of the Literature

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Case report

Keywords: subclavian steal syndrome; subclavian artery aneurysm; ultrasonography

Posted Date: February 6th, 2020

DOI: https://doi.org/10.21203/rs.2.22738/v1

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Abstract

Background "Stealing" of the subclavian artery is a phenomenon where proximal subclavian artery stenosis or occlusion causes retrograde blood flow in the ipsilateral vertebral artery. This, combined with ischemic symptoms of vertebrobasilar arteries or ipsilateral upper limb artery, is called subclavian steal syndrome. Sometimes, subclavian steal syndrome can occur in patients without subclavian artery stenosis, such as in patients with high-flow arteriovenous dialysis fistulas or subclavian artery aneurysms. Interestingly, compared with classic atherosclerotic subclavian steal syndrome, the former patient is more prone to symptoms like paroxysmal vertigo, drop attacks, diplopia, and arm claudication. Case Presentation In our case, we present a 35-year-old male who suffered from paroxysmal vertigo. Clinical physical examination did not reveal any substantial evidence of proximal subclavian artery stenosis. Transcranial Doppler and ultrasonography revealed an abnormal blood flow pattern in the right vertebral artery, consistent with partial subclavian steal syndrome. Ultrasonography and computed tomography angiography showed no stenosis in the right subclavian artery and three aneurysms in the proximal segment. The hypoplastic vertebral artery originated from the third aneurysm. Conclusions There are few reports of subclavian steal syndrome caused by subclavian aneurysms in the literature; our case is the first to report partial subclavian steal syndrome caused by multiple aneurysms. Following a literature review, combined with our analysis of this case, we suggest that subclavian artery aneurysm which induced subclavian steal syndrome is an often unnoticed cause of vertigo.

Background

The pathophysiological changes of subclavian steal syndrome (SSS) are stenosis or occlusion of the subclavian artery or innominate artery, which are proximal to the origin of the vertebral artery (VA). [1] The most common cause is the atherosclerotic stenosis revealed by ultrasonography.[2–3] Subclavian artery aneurysm (SAA) is a rare type of peripheral vascular aneurysm that manifests various clinical symptoms, including compression syndrome, thrombosis, or rupture, with the risk of distal embolism and stroke.[4] There are few reports of partial subclavian steal syndrome caused by subclavian aneurysms. An extensive literature review revealed 4 case reports that described SSS without stenosis[5 – 8]; the steal was attributed to the SAA (Table 1).

Table 1 Review of the literature about subclavian artery aneurysm-induced subclavian steal syndrome

Author	Otto R[5]	Mingyu X[6] 2005	Youhe J[7]	Buljan K[8]	**Zhe L**
Date	1986		2012	2014	**2019**
Gender, Age	male , 47	female , 77	female , 57	male , 55	**male, 35**
Location, Quantity	Right, 1	Righ, 1	Right, 1	Right , 1	**Right, 3**
Size	6.0x6.0cm	1.39x1.39cm	1.75x1.95cm	1.56 cm	**2.5cmx2.3cm, 1.4cmx1.1cm, 1.1cmx0.9cm**
Imaging examination	CT , DSA	TCD , US	TCD , US , CTA	US , CTA	**TCD, US, CTA**
Degree of SSS	II or III	I	I	I	**II**
VA diameter	Unknown	1.9mm	1.7mm	2mm	**1.9mm**
Point of VA origin	Unknown	Aneurysm	Aneurysm	Aneurysm	**Aneurysm**

CT, Computed Tomography; DSA, Digital subtraction angiography; TCD, Transcranial Doppler; US, ultrasonography; CTA, Computed Tomography Angiography; SSS, subclavian steal syndrome; VA, vertebral artery; Bold part described our case.

Case Presentation

35-year-old male complained of sporadic dizziness for about 1 year and went to our hospital. The patient past history included: no recent history of head and neck trauma; denial of hypertension, diabetes and any cardiovascular disease; no history of alcohol consumption and smoking. Physical examination revealed normal cardiopulmonary function, neck was not touching lymph nodes, bilateral clavicle were not touching mass, body temperature 36.8 C, pulse 68 times/min, breathing 16 times/min, and left and right upper limb blood pressure 125/90 mmHg and 110/80 mmHg, respectively.

Transcranial Doppler (TCD) examination revealed a retrograde spectrum in the V4 segment of VA while late systole(Fig. 1).The remaining intracranial arteries were normal. An ultrasound examination of the extracranial cervical vessels revealed normal bilateral carotid arteries and left subclavian artery without plaque or thrombus. (7.5 Mhz linear array probe, Toshiba Aplio 5000). A retrograde spectrum in the V2 segment of VA while mid-systole was detected via ultrasonography(Fig. 2). During the right VA Doppler scan, a full cardiac cycle inversion spectrum was found while the patient kept fist-releasing with his right hand (Fig. 3).

The lumen of the innominate artery and the proximal right subclavian artery was unobstructed with the normal intima-media thickness (Fig. 4). An aneurysmal echo (2.5 × 2.3 cm) in the middle of subclavian artery was detected with a swirling blood flow pattern (Fig. 5A). Another aneurysmal echo (1.4 × 1.1 cm) was detected at the outlet of the first aneurysm with a swirling blood flow pattern (Fig. 5B). Below the

aneurysm, the subclavian artery was tortuous with a narrow diameter. Another aneurysmal echo (1.1 × 0.9 cm) was detected, where the right VA originated (Fig. 5C, 5D). We found an abnormal spectrum of low-speed and a low pulsation index in the right radial artery. Subsequently, CTA revealed three aneurysms of the right subclavian artery and confirmed the US diagnosis (Fig. 6) that the right vertebral artery originated from the third aneurysm.

Disscusion

We reported a rare case of SSS without stenosis induced by multiple SAA that was diagnosed via comprehensive head and neck ultrasonography. The report was consented with the patient. The pathophysiological changes of SSS are stenosis or occlusion of the subclavian artery proximal to the origin of the VA. The most common cause is the atherosclerotic stenosis. The hemodynamic mechanism of SSS is the abnormal pressure gradient between the bilateral vertebral arteries due to the lesion of subclavian. [1] In general, a duplex ultrasound can be used to observe the characteristic spectrum of alternating directions in VA, due to SSS. Previous studies have shown that SSS is often associated with the degree of subclavian artery stenosis proximal to the VA to some extent.[3]

We found that the pathophysiological mechanisms of vertebral artery hemodynamics in SAA patients and subclavian artery stenosis patients were similar. Although uncommon, multiple SAA on the right is the cause of vertebral arterial blood reflux in our case. The origin of VA in our case was located on the aneurysm. SSS may be explained by Bernoulli's equation and Venturi-style management theory.[9] According to the Bernoulli equation, the potential energy of Newton's liquid is inversely proportional to the kinetic energy. As the velocity (i.e., kinetic energy) increases, the pressure (i.e., potential energy) decreases. Vice versa,the pressure inside the aneurysm decreases, and the blood flow speed inside the aneurysm will inevitably accelerate. According to the Venturi tube principle, the acceleration of blood flow at the vertebral artery opening will cause the reduction of pressure at the same point, and then the aneurysm originating from the VA will "suck" the blood from the basilar artery or the contralateral VA，which can cause vertebrobasilar insufficiency. Depending on the severity of SSS, there will be a variety of ischemic symptoms in the posterior circulation, which might be the source of occasional dizziness of our patient.

The symptoms of different SAAs are variable. [10] Nearly up to a third of patients with SAA were asymptomatic. [11] Saliou and his team explained that intrathoracic SAA is largely asymptomatic and difficult to detect on physical examination, and that symptoms may be directly related to the size of the aneurysm.[12] However, upon reviewing previous studies, we found that the diagnosis of SAA was mostly achieved through advanced imaging examination, which gave the message of the location and size of SAA.[10] However, most of these examinations could not provide hemodynamic parameters, such as CT, CTA, and MRA. Few studies analyzed the possible hemodynamic changes in the VA of patients with SAA. Ultrasonography is a reliable method for the evaluation of the VA hemodynamics. In general, it might be the only diagnostic tool sensitive enough to detect SSS I .[3] In our case ,during the doppler detection of

right VA, the spectrum changed from a mid-systolic inversion to a full cardiac cycle inversion while the patient kept fist-releasing with his right hand (Fig. 3).Through this clinical trials, the increased demand of blood in the right upper extremity resulted in the patient moving from SSS II to SSS III; the continuous variation in the form of vertebral artery spectrum was recorded vividly by VA ultrasonography. These advantages of ultrasonography lead us to recommend this method for the early and accurate diagnosis and follow-up of low-pressure lesions in the subclavian artery.

Compared with the aforementioned cases (Table 1), [5–8] the patient we reported were younger, with no obvious signs of arteriosclerosis, which suggests the right SAA was more likely to be congenital. In addition, our patient had multiple SAAs, and as the number increased, so did the severity of SSS. In the case of Otto, [5] the aneurysm was substantially large, and the patient's degree of SSS was also aggravated compare with other reports. Therefore, we speculated that the number and size of aneurysms might be responsible for the severity of SSS. In summary, our case provide increasing evidence that SAA is a possible underlying cause of SSS, suggesting that low-pressure lesions in the subclavian artery are a potential cause of vertebrobasilar insufficiency.

Conclusions

We conclude that if a subclavian aneurysm is found, vertebral artery ultrasound examination should be used to observe whether there is retrograde blood flow in the ipsilateral vertebral artery to explain patients' posterior circulation ischemia symptoms. We suggest that subclavian artery aneurysm-induced subclavian steal syndrome is an often unnoticed cause of vertigo. Thus, clinicians treating young patients who suffer from vertebrobasilar insufficiency should consider low-pressure lesions in the subclavian artery as a possible cause, and give patients a comprehensive ultrasonography of the head and neck vessels and the subclavian artery.

List Of Abbreviations

Subclavian steal syndrome	SSS
Subclavian artery aneurysm	SAA
Vertebral artery	VA
Transcranial Doppler	TCD
Computed Tomography	CT
Digital subtraction angiography	DSA
Computed Tomography Angiography	CTA
Magnetic resonance angiography	MRA

Declarations

Ethics approval and consent to participate

Not applicable for ethics approval. The case report was consented with the patient.

Consent for publication

Not applicable.

Availability of data and materials

Not applicable.

Competing interests

The authors declare that they have no competing interests.

Funding

Not applicable.

Authors' contributions

"QBL,CYM,YHJ,XW analyzed and interpreted the patient ultrasonic data regarding the subclavian steal syndrome disease. ZL performed the ultrasound examination of the patient, and was a major contributor in writing the manuscript. All authors read and approved the final manuscript."

Acknowledgements

Not applicable.

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Figures

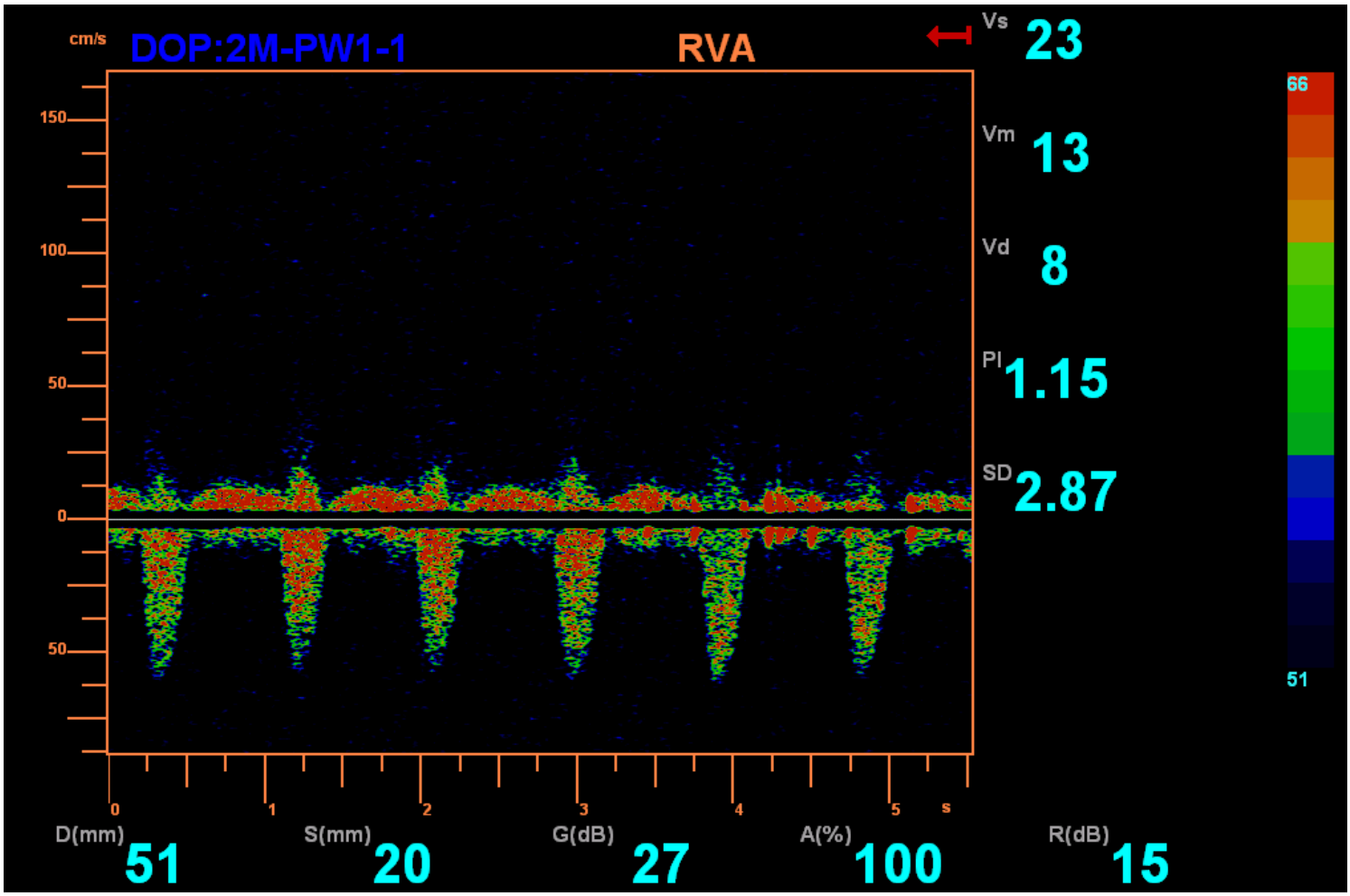


Figure 2

The right vertebral artery spectrum of SSS II.



Figure 4

Sonography of right subclavian artery. A Two-dimensional ultrasound revealed a normal pattern in the V0-V1 segment of the right VA. B CDFI revealed a normal color pattern in the V0-V1 segment of the right VA. C Duplex ultrasound revealed a retrograde spectrum in the V2 segment of VA while mid-systole and returned to normal during diastole.

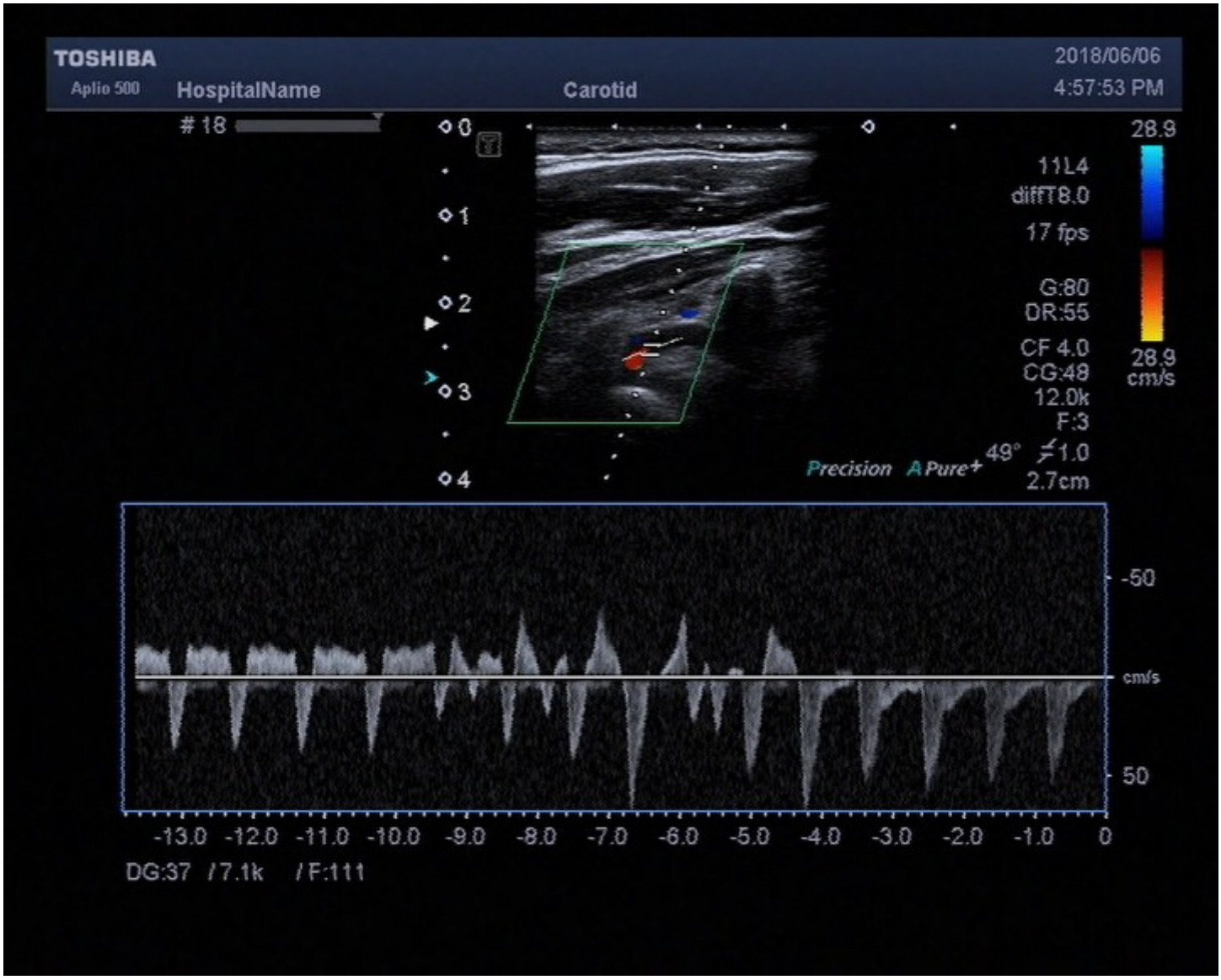


Figure 6

The spectrum of right VA changed from SSS II to SSS III, while the patient kept fist-releasing with his right hand.

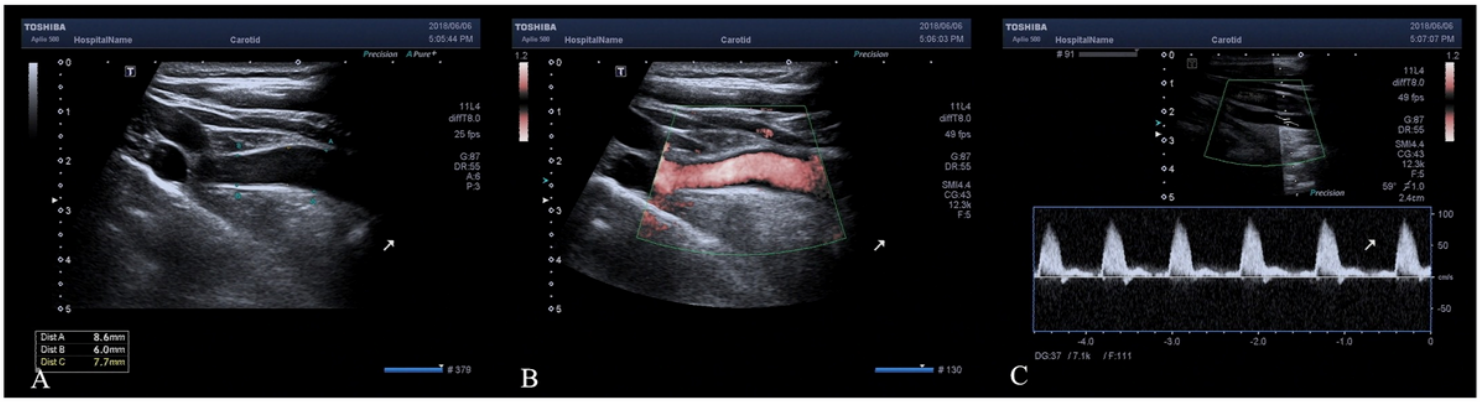


Figure 8

Sonography of proximal right subclavian artery A lumen of the innominate artery and the proximal right subclavian artery was unobstructed with normal intima-media thickness. B CDFI revealed a normal color pattern of proximal right subclavian artery C The proximal segment of right subclavian artery with a normal artery spectrum

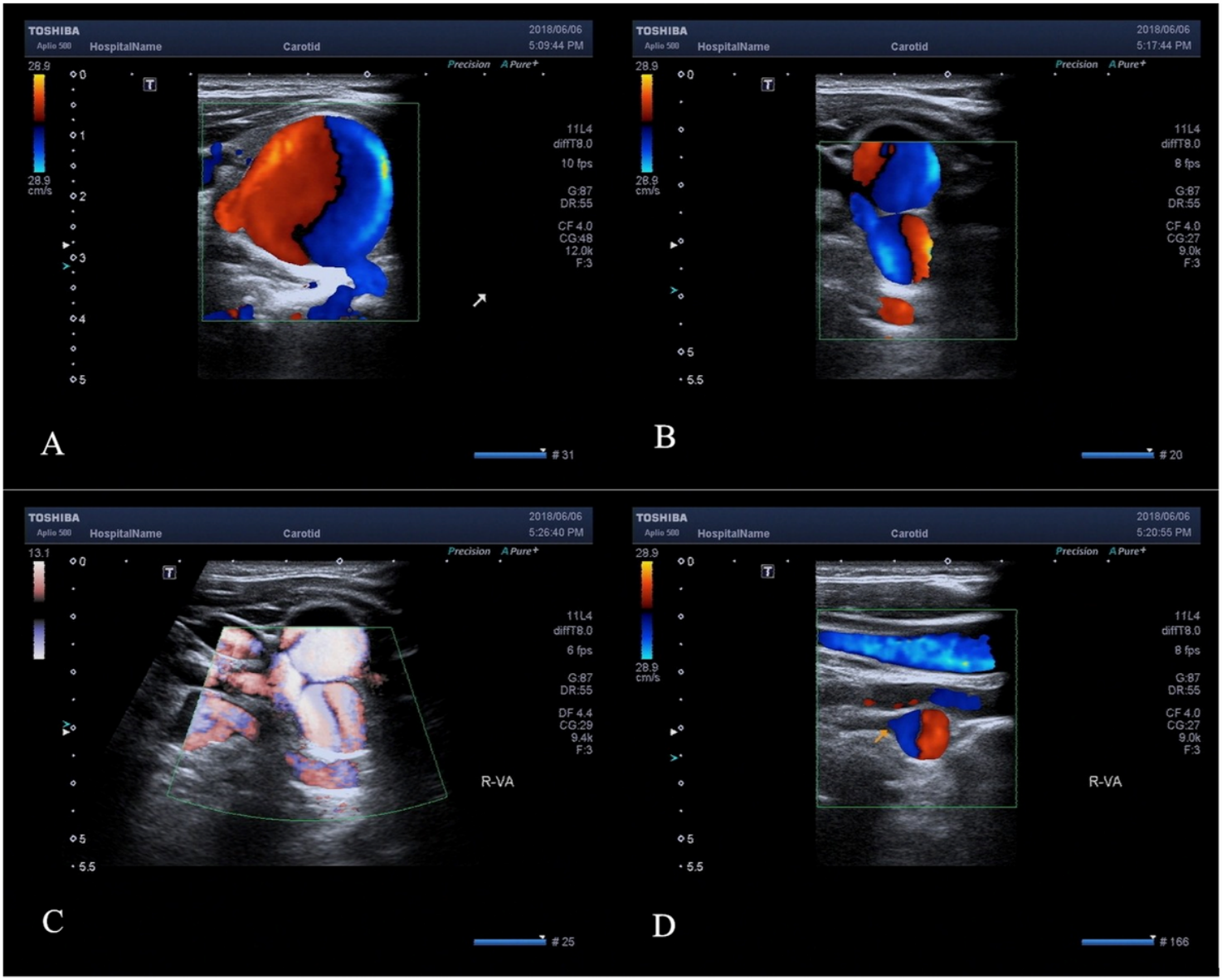


Figure 9

Three aneurysms of the right subclavian artery in sonography. A The first aneurysm, CDFI like a logo of Pepsi Cola. B The second aneurysm, CDFI like a logo of Pepsi Cola. C,D The third aneurysm, CDFI like a logo of Pepsi Cola. The right vertebral artery originated from the third aneurysm.

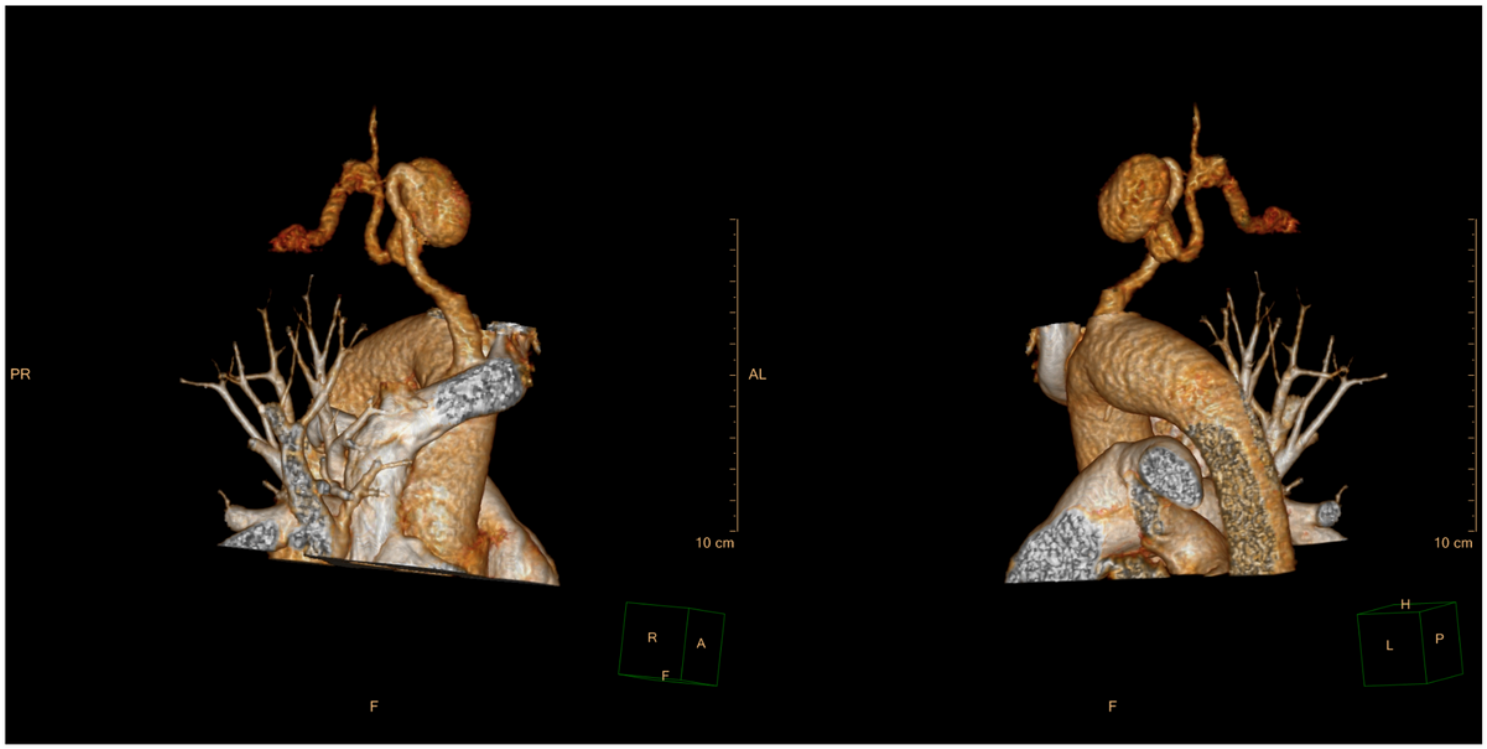


Figure 11

CTA revealed three aneurysms of the right subclavian artery and right vertebral artery originated from the central of the third aneurysm.