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**Title**

# Presurgical assessment of flow variability in an azygos vein aneurysm using 4D-flow MRI.

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**Keywords**

4D-flow MRI; azygos vein aneurysm

**Abstract**

Azygos vein aneurysm (AVA) is necessary to prevent pulmonary embolism due to the outflow of a thrombus or rupture of the aneurysm. However, there is no established modality to assess the properties of AVA. Time-resolved three-dimensional phase-contrast magnetic resonance imaging (4D-flow MRI) has been used to examine the hemodynamics in various fields. We report a case of AVA to evaluate the flow variability and adhesions of surrounding tissues using 4D-flow MRI. The findings of the study suggested aneurysm turbulence and the absence of thrombi. The cine image, which showed a sliding wall synchronized to the heartbeat, indicated no adhesion to the superior vena cava. Based on these results, the thoracoscopic approach was deemed possible preoperatively. Thoracoscopic AVA resection was performed, and the postoperative course was uneventful. This study documented the utility of 4D-flow MRI for a detailed evaluation of AVA.

## Introduction

Azygos vein aneurysm (AVA) is a rare disease that may present with hemoptysis, chest pain, and palpitation. However, no symptoms are observed in most cases. Appropriate treatment of AVA is essential because it leads to pulmonary embolism due to thrombus outflow or possible rupture of the aneurysm. Since these events can also occur during surgery, preoperative evaluation of the status of thrombus in the aneurysm and adhesions of surrounding tissues is important for surgical risk assessment. Time-resolved three-dimensional phase-contrast magnetic resonance imaging (4D-flow MRI) has been applied in various fields, such as the cardiovascular and neurovascular regions. It visualizes the hemodynamics and estimates the prognosis of aneurysms [1, 2]. 4D-flow MRI also determines thrombus formation in the left atrium after a left upper lobectomy in thoracic surgery [3]. We report the application of 4D-flow MRI to evaluate the flow variability and surrounding in case of AVA.

## Case

A 65-year-old woman with a history of asthma presented with an atypical enlargement of the right mediastinal contour on chest X-ray imaging during a medical checkout. She had no chief complaints, and no abnormalities were found in the blood tests, including a coagulation study. Contrast-enhanced computed tomography (CT) revealed idiopathic saccular AVA (defined as an eccentric focal dilatation bulging out from a part of the azygos vein [4]), measuring 60 × 30 × 55 mm. She was referred to our hospital for surgery. However, the AVA had expanded to 70 × 40 × 60 mm by the time of referral, which was about 2 months after the last evaluation at the previous center. The mediastinal side of the AVA was adjacent to the superior vena cava (SVC), and the SVC was slightly compressed by the AVA. Moreover, the boundary between the AVA and the SVC was unclear. Thus, adhesions could not be denied on dynamic CT (Fig. a-c). To address this, 4D-flow MRI was conducted. The streamline images of 4D-flow MRI showed a slow turbulent blood flow within the AVA (Fig. f, g, and Online Resource). In addition, T1 and T2 weighted black-blood images showed a heterogeneous signal intensity inside the AVA (Fig. d). The AVA's internal turbulence blood flow suggested that the heterogeneous signal intensity in T1 and T2 weighted black-blood images reflected turbulence rather than thrombus. Based on this information, the blood results, and the normal coagulation, there were likely no thrombi. The adhesions between the AVA and the SVCs were also excluded because the cine images of the magnetic resonance imaging (MRI) showed the sliding mobility synchronized with the heartbeat (Fig. e, and Online Resource). These results supported the feasibility of the thoracoscopic approach (no need for open thoracotomy). Under 2-port thoracoscopic surgery, the aneurysm wall was significantly thin, that even a slight dissection caused oozing. Therefore, careful dissection of the mediastinal pleura was required. The AVA was adjacent to the SVC and mediastinal pleura, but no adhesions were observed (Fig. h and i). To ensure safety, the central side of the SVC was encircled by vessel loops. Both the peripheral and central sides of the AVA were dissected with a surgical stapler and the AVA was removed safely. No thrombus was observed in the resected specimen. The postoperative course was uneventful, and the patient was discharged eight days after the operation. Since then, no aneurysm recurrence has been observed.

**Discussion**

AVA is a rare disease, whose etiology can be divided into idiopathic, secondary, and traumatic [5]. Although some studies have reported that anatomical and embryological frailty were involved in the formation of idiopathic AVA, the exact reason remains unknown [6, 7]. Causes of secondary AVA include heart failure, portal hypertension, pregnancy, congenital anomalies of the vena cava, and acquired obstruction associated with tumors or thrombosis [5, 8]. In terms of treatment, Ko et al. [4] reported that saccular AVAs require surgical intervention due to the greater frequency of chest symptoms and intralesional thrombosis (all 4 saccular AVAs in their study were thrombosed). They noted that if thrombosis is detected, emergency surgery or oral anticoagulant treatment should be attempted first, however, there is no consensus on this yet [4, 5]. Meanwhile, Ko et al. [4] also reported that fusiform AVAs (defined as circumferential short-segment spindle-shaped dilatation of the azygos vein) should only be carefully followed-up, because all 6 fusiform AVAs in this study were asymptomatic and not thrombosed. Studies on surgical interventions for AVA have reported the utility of the thoracoscopic approach [5]. However, massive intraoperative bleeding has been reported during thoracoscopic surgery [9]. Furthermore, some reports have associated the risk of pulmonary thromboembolism with thrombus migration [4, 5]. Thus, the status of the thrombus in the AVA and the presence of adhesions within the surrounding tissues due to inflammation [4, 10], which predisposes to massive bleeding, are important considerations when determining the indications for the thoracoscopic approach [10]. Although, contrast-enhanced CT and MRI have been used as the primary modalities [4, 5], they may yield images that resemble solid masses or a heterogeneous contrast enhancement, especially if the blood flow inside the AVA is slow or if complete thrombosis exists. This can lead to misdiagnoses, such as thymoma, lymphoma, and neurogenic tumor, making an evaluation of thrombus impossible [4-6]. In addition, the relationship with surrounding tissues is often vague, as in the present case, therefore, the evaluation of adhesion seemed difficult. In 4D-flow MRI, analyzed numerical and visual information, such as the blood flow velocity/direction, are obtained in addition to the information obtained by conventional MRI. Even if CT or MRI cannot distinguish solid masses, the diagnosis is made accurate by findings suggestive of turbulent blood flow within the AVA in 4D-flow MRI, as seen in the present case. In cases where a heterogeneous signal intensity is observed in T1 and T2 weighted black-blood MRI images, and the 4D-flow MRI findings suggest AVA turbulence, a thrombus is less likely than turbulence. This impression is more reliable when the blood test reveals a normal coagulation system. Moreover, the adhesion between the AVA and the surrounding tissues is preoperatively diagnosed based on the sliding motion observed, depending on the time axis in the cine images of the MRI. In the present case, the adhesion between the AVA and the SVC, which was unclear on contrast-enhanced CT, was clearly appreciated in the MRI cine images. Thus, the surgical approach was determined preoperatively. A disadvantage of 4D-flow MRI may be the prolonged time required for images acquisition, depending on heart rate, breathing compensation efficiency, and applied sequence [1], it takes an additional 5-20 min longer than the imaging time required for conventional MRI. Another disadvantage is that it requires a special software for analyzing the images. In addition, it is necessary to set velocity encoding (VENC) in 4D-flow MRI. If the VENC settings favor the visualization of large blood vessels, communicating with the AVA, the blood flow of the AVA is challenging to evaluate when the velocity is low. Despite these challenges, 4D-Flow MRI can be applied to a wider range of AVA patients without contrast media and radiation exposure. Therefore, 4D-flow MRI is a valuable option for the evaluation of AVA, especially in patients with renal dysfunction and pregnant.

**Conclusion**

4D-flow MRI was useful for AVA evaluation. More AVA case studies are needed to confirm the utility of the 4D-flow MRI in clinical practice.

## Disclosure statement

There is no conflict of interest.

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## Figure Legends

**Fig.**

Differences in the diagnostic capability for azygos vein aneurysms (AVAs) between contrast-enhanced computed tomography, three-dimensional (3D) reconstruction of azygos vein aneurysm (a-c), and four-dimensional (4D) flow magnetic resonance imaging (MRI) (d-f). And Intraoperative thoracoscopic imaging (h, i).

**(a)** Early phase axial view of the 70 × 40 × 60 mm AVA. It was proximal to the superior vena cava (SVC), and the AVA's internal contrast effect is heterogeneous.

**(b)** Sagittal view, dynamic phase, showing that the aneurysm was formed in a shape that protruded mainly into the cranial direction from the original azygos vein arch and was judged to be a saccular AVA.

**(c)** In volume rendering, it was impossible to determine whether adhesions existed between the AVA and the SVC.

**(d)** T1-weighted black-blood image, axial view of MRI. The boundary between the SVC and the AVA can be determined, and the signal inside the AVA is heterogeneous.

**(e)** Cine image, sagittal view. It showed sliding mobility between the AVA and the SVC synchronized to the heartbeat.

**(f, g)** Two streamline images of 4D-flow MRI in the venous system with different phases of the cardiac cycle. A slightly slow turbulent flow was observed.

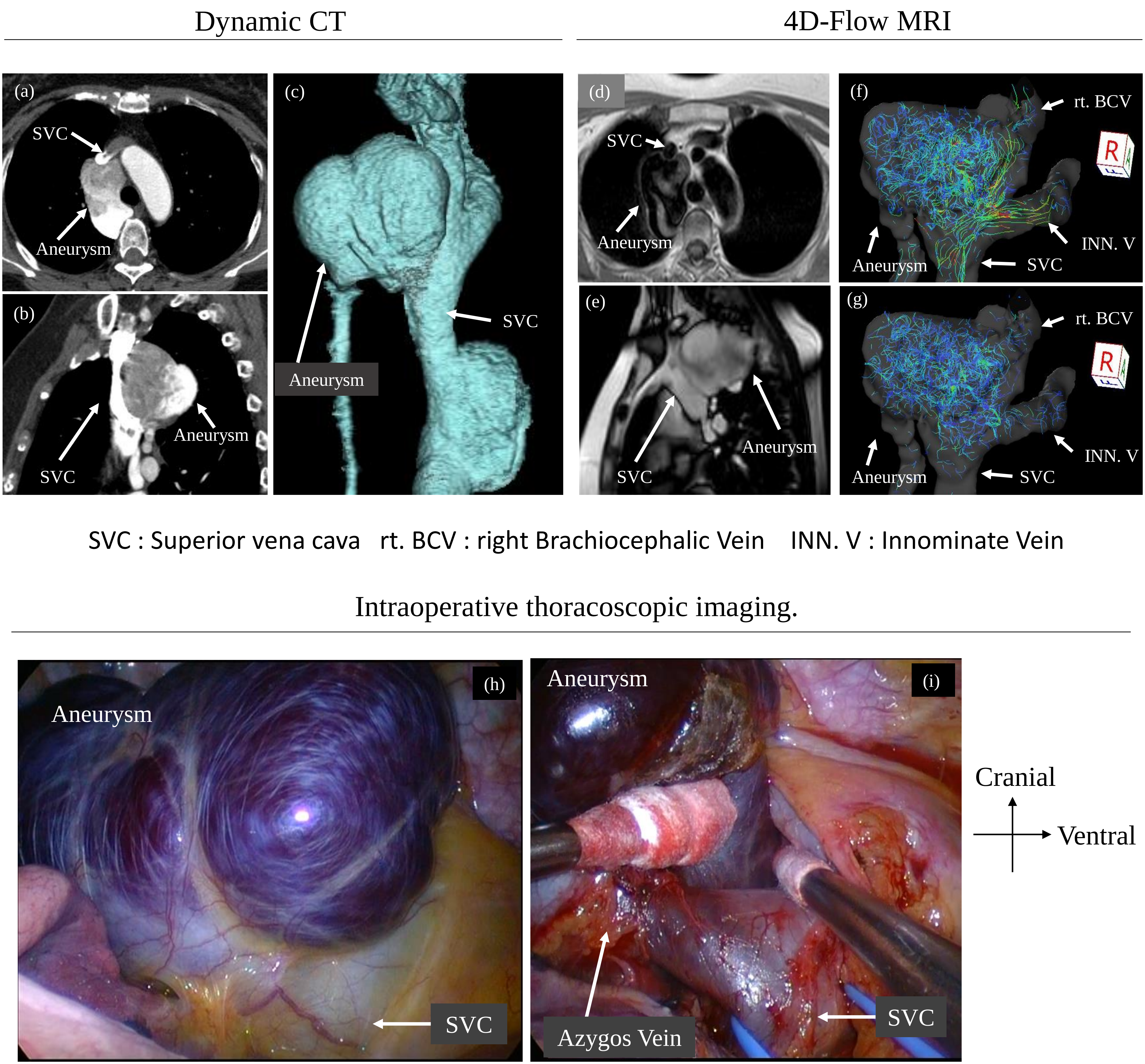
**(h)** The AVA before dissection.

**(i)** The AVA and the SVC were adjacent to each other within the mediastinal pleura, but no adhesions were observed

**Online Resource**

Findings of cine images (0:00-0:16), and 4D-flow MRI: Analyzed movie of the blood flow velocity/direction (0:17-0:27).

The cine MRI images showed sliding mobility between the AVA and the SVC synchronized to the heartbeat. In the analyzed movie of the blood flow velocity/direction, a slight change in the color of the streamline over time was observed, and it was suggested that there was internal turbulent blood flow.


Dynamic CT
4D-Flow MRI
(a)
SVC
Aneurysm
(b)
Aneurysm
SVC
(c)
Aneurysm
SVC
(d)
SVC
Aneurysm
(e)
SVC
Aneurysm
(f)
rt. BCV
INN. V
Aneurysm
SVC
(g)
rt. BCV
INN. V
Aneurysm
SVC
SVC : Superior vena cava rt. BCV : right Brachiocephalic Vein INN. V : Innominate Vein
Intraoperative thoracoscopic imaging.
(h)
Aneurysm
SVC
(i)
Aneurysm
Azygos Vein
SVC
Cranial
Ventral