



HOKKAIDO UNIVERSITY

Title	Presurgical assessment of flow variability in an azygos vein aneurysm using 4D-flow MRI
Author(s)	Takuya, Ikushima; Hideki, Ujiie; Satonori, Tsuneta et al.
Citation	General Thoracic and Cardiovascular Surgery, 70(7), 673-676 https://doi.org/10.1007/s11748-022-01813-7
Issue Date	2022-07
Doc URL	https://hdl.handle.net/2115/90106
Rights	This version of the article has been accepted for publication, after peer review (when applicable) and is subject to Springer Nature's AM terms of use, but is not the Version of Record and does not reflect post-acceptance improvements, or any corrections. The Version of Record is available online at: http://dx.doi.org/10.1007/s11748-022-01813-7
Type	journal article
File Information	GTCS 70 673-676.pdf



1 **Title**

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

3

4 **Authors:**

5 Takuya Ikushima, MD¹, Hideki Ujiie, MD, PhD, FACS¹, Satonori Tsuneta, MD, PhD², Ryohei

6 Chiba, MD¹, Yukiko Tabata, MD, PhD¹, Aki Fujiwara-Kuroda, MD, PhD¹, Yasuhiro Hida, MD,

7 PhD¹, Kichizo Kaga, MD, PhD¹, Satoru Wakasa, MD, PhD¹, and Tatsuya Kato, MD, PhD¹

8

9 **Institutions and Affiliations**

10 ¹ Department of Cardiovascular and Thoracic Surgery, Faculty of Medicine and Graduate

11 School of Medicine, Hokkaido University, Sapporo, Hokkaido, Japan

12 ² Department of Diagnostic and Interventional Radiology, Hokkaido University Hospital, Sapporo,

13 Hokkaido, Japan

14

15 **Corresponding Author**

16 Dr. Tatsuya Kato

17 Department of Cardiovascular and Thoracic Surgery, Faculty of Medicine and Graduate School

18 of Medicine, Hokkaido University

1 Kita 15, Nishi 7, Kita-ku, Sapporo, Hokkaido 060-8638,

2 Japan

3 Tel.: +81-11-706-6042; Fax: +81-11-706-7612

4 Email: katotatu@huhp.hokudai.ac.jp

5

6 **Meeting presentation**

7 The 74th Annual Scientific Meeting of the Japanese Association for Thoracic Surgery

8

9 **Keywords**

10 4D-flow MRI; azygos vein aneurysm

11

12

13

14

15

16

17

18

1 **Abstract**

2 Azygos vein aneurysm (AVA) is necessary to prevent pulmonary embolism due to the outflow of
3 a thrombus or rupture of the aneurysm. However, there is no established modality to assess the
4 properties of AVA. Time-resolved three-dimensional phase-contrast magnetic resonance imaging
5 (4D-flow MRI) has been used to examine the hemodynamics in various fields. We report a case
6 of AVA to evaluate the flow variability and adhesions of surrounding tissues using 4D-flow MRI.
7 The findings of the study suggested aneurysm turbulence and the absence of thrombi. The cine
8 image, which showed a sliding wall synchronized to the heartbeat, indicated no adhesion to the
9 superior vena cava. Based on these results, the thoracoscopic approach was deemed possible
10 preoperatively. Thoracoscopic AVA resection was performed, and the postoperative course was
11 uneventful. This study documented the utility of 4D-flow MRI for a detailed evaluation of AVA.

12

13

14

15

16

17

18

1 **Introduction**

2 Azygos vein aneurysm (AVA) is a rare disease that may present with hemoptysis, chest pain, and
3 palpitation. However, no symptoms are observed in most cases. Appropriate treatment of AVA is
4 essential because it leads to pulmonary embolism due to thrombus outflow or possible rupture of
5 the aneurysm. Since these events can also occur during surgery, preoperative evaluation of the
6 status of thrombus in the aneurysm and adhesions of surrounding tissues is important for surgical
7 risk assessment. Time-resolved three-dimensional phase-contrast magnetic resonance imaging
8 (4D-flow MRI) has been applied in various fields, such as the cardiovascular and neurovascular
9 regions. It visualizes the hemodynamics and estimates the prognosis of aneurysms [1, 2]. 4D-flow
10 MRI also determines thrombus formation in the left atrium after a left upper lobectomy in thoracic
11 surgery [3]. We report the application of 4D-flow MRI to evaluate the flow variability and
12 surrounding in case of AVA.

13

14 **Case**

15 A 65-year-old woman with a history of asthma presented with an atypical enlargement of the right
16 mediastinal contour on chest X-ray imaging during a medical checkout. She had no chief
17 complaints, and no abnormalities were found in the blood tests, including a coagulation study.
18 Contrast-enhanced computed tomography (CT) revealed idiopathic saccular AVA (defined as an

1 eccentric focal dilatation bulging out from a part of the azygos vein [4]), measuring $60 \times 30 \times 55$
2 mm. She was referred to our hospital for surgery. However, the AVA had expanded to $70 \times 40 \times$
3 60 mm by the time of referral, which was about 2 months after the last evaluation at the previous
4 center. The mediastinal side of the AVA was adjacent to the superior vena cava (SVC), and the
5 SVC was slightly compressed by the AVA. Moreover, the boundary between the AVA and the SVC
6 was unclear. Thus, adhesions could not be denied on dynamic CT (Fig. a-c). To address this, 4D-
7 flow MRI was conducted. The streamline images of 4D-flow MRI showed a slow turbulent blood
8 flow within the AVA (Fig. f, g, and Online Resource). In addition, T1 and T2 weighted black-
9 blood images showed a heterogeneous signal intensity inside the AVA (Fig. d). The AVA's internal
10 turbulence blood flow suggested that the heterogeneous signal intensity in T1 and T2 weighted
11 black-blood images reflected turbulence rather than thrombus. Based on this information, the
12 blood results, and the normal coagulation, there were likely no thrombi. The adhesions between
13 the AVA and the SVCs were also excluded because the cine images of the magnetic resonance
14 imaging (MRI) showed the sliding mobility synchronized with the heartbeat (Fig. e, and Online
15 Resource). These results supported the feasibility of the thoracoscopic approach (no need for open
16 thoracotomy). Under 2-port thoracoscopic surgery, the aneurysm wall was significantly thin, that
17 even a slight dissection caused oozing. Therefore, careful dissection of the mediastinal pleura was
18 required. The AVA was adjacent to the SVC and mediastinal pleura, but no adhesions were

1 observed (Fig. h and i). To ensure safety, the central side of the SVC was encircled by vessel loops.
2 Both the peripheral and central sides of the AVA were dissected with a surgical stapler and the
3 AVA was removed safely. No thrombus was observed in the resected specimen. The postoperative
4 course was uneventful, and the patient was discharged eight days after the operation. Since then,
5 no aneurysm recurrence has been observed.

6

7 **Discussion**

8 AVA is a rare disease, whose etiology can be divided into idiopathic, secondary, and traumatic [5].
9 Although some studies have reported that anatomical and embryological frailty were involved in
10 the formation of idiopathic AVA, the exact reason remains unknown [6, 7]. Causes of secondary
11 AVA include heart failure, portal hypertension, pregnancy, congenital anomalies of the vena cava,
12 and acquired obstruction associated with tumors or thrombosis [5, 8]. In terms of treatment, Ko
13 et al. [4] reported that saccular AVAs require surgical intervention due to the greater frequency of
14 chest symptoms and intralesional thrombosis (all 4 saccular AVAs in their study were thrombosed).
15 They noted that if thrombosis is detected, emergency surgery or oral anticoagulant treatment
16 should be attempted first, however, there is no consensus on this yet [4, 5]. Meanwhile, Ko et al.
17 [4] also reported that fusiform AVAs (defined as circumferential short-segment spindle-shaped
18 dilatation of the azygos vein) should only be carefully followed-up, because all 6 fusiform AVAs

1 in this study were asymptomatic and not thrombosed. Studies on surgical interventions for AVA
2 have reported the utility of the thoracoscopic approach [5]. However, massive intraoperative
3 bleeding has been reported during thoracoscopic surgery [9]. Furthermore, some reports have
4 associated the risk of pulmonary thromboembolism with thrombus migration [4, 5]. Thus, the status
5 of the thrombus in the AVA and the presence of adhesions within the surrounding tissues due to
6 inflammation [4, 10], which predisposes to massive bleeding, are important considerations when
7 determining the indications for the thoracoscopic approach [10]. Although, contrast-enhanced CT
8 and MRI have been used as the primary modalities [4, 5], they may yield images that resemble
9 solid masses or a heterogeneous contrast enhancement, especially if the blood flow inside the
10 AVA is slow or if complete thrombosis exists. This can lead to misdiagnoses, such as thymoma,
11 lymphoma, and neurogenic tumor, making an evaluation of thrombus impossible [4-6]. In
12 addition, the relationship with surrounding tissues is often vague, as in the present case, therefore,
13 the evaluation of adhesion seemed difficult. In 4D-flow MRI, analyzed numerical and visual
14 information, such as the blood flow velocity/direction, are obtained in addition to the information
15 obtained by conventional MRI. Even if CT or MRI cannot distinguish solid masses, the diagnosis
16 is made accurate by findings suggestive of turbulent blood flow within the AVA in 4D-flow MRI,
17 as seen in the present case. In cases where a heterogeneous signal intensity is observed in T1 and
18 T2 weighted black-blood MRI images, and the 4D-flow MRI findings suggest AVA turbulence, a

1 thrombus is less likely than turbulence. This impression is more reliable when the blood test
2 reveals a normal coagulation system. Moreover, the adhesion between the AVA and the
3 surrounding tissues is preoperatively diagnosed based on the sliding motion observed, depending
4 on the time axis in the cine images of the MRI. In the present case, the adhesion between the AVA
5 and the SVC, which was unclear on contrast-enhanced CT, was clearly appreciated in the MRI
6 cine images. Thus, the surgical approach was determined preoperatively. A disadvantage of 4D-
7 flow MRI may be the prolonged time required for images acquisition, depending on heart rate,
8 breathing compensation efficiency, and applied sequence [1], it takes an additional 5-20 min
9 longer than the imaging time required for conventional MRI. Another disadvantage is that it
10 requires a special software for analyzing the images. In addition, it is necessary to set velocity
11 encoding (VENC) in 4D-flow MRI. If the VENC settings favor the visualization of large blood
12 vessels, communicating with the AVA, the blood flow of the AVA is challenging to evaluate when
13 the velocity is low. Despite these challenges, 4D-Flow MRI can be applied to a wider range of
14 AVA patients without contrast media and radiation exposure. Therefore, 4D-flow MRI is a
15 valuable option for the evaluation of AVA, especially in patients with renal dysfunction and
16 pregnant.

18 **Conclusion**

1 4D-flow MRI was useful for AVA evaluation. More AVA case studies are needed to confirm the
2 utility of the 4D-flow MRI in clinical practice.

3

4 **Disclosure statement**

5 There is no conflict of interest.

6

7 **Acknowledgements**

8 We would like to thank Editage (www.editage.com) for English language editing.

9

10

11

12

13

14

15

16

17

18

1 **References**

- 2 1. Markl M, Frydrychowicz A, Kozerke S, Hope M, Wieben O. 4D flow MRI. *J Magn Reson*
3 Imaging. 2012;36:1015–36.
- 4 2. Sträter A, Huber A, Rudolph J, Berndt M, Rasper M, Rummeny EJ, et al. 4D-flow MRI:
5 Technique and applications. *RoFo*. 2018;190:1025–35.
- 6 3. Matsumoto M, Takegahara K, Inoue T, Nakaza M, Sekine T, Usuda J. 4D flow MR
7 imaging reveals a decrease of left atrial blood flow in a patient with cardioembolic cerebral
8 infarction after pulmonary left upper lobectomy. *Magn Reson Med Sci*. 2020;19:290–3.
- 9 4. Ko SF, Huang CC, Lin JW, Lu HI, Kung CT, Ng SH, et al. Imaging features and outcomes
10 in 10 cases of idiopathic azygos vein aneurysm. *Ann Thorac Surg*. 2014;97:873–8.
- 11 5. Kreibich M, Siepe M, Grohmann J, Pache G, Beyersdorf F. Aneurysms of the azygos vein.
12 *J Vasc Surg Venous Lymphat Disord*. 2017;5:576–86.
- 13 6. Gallego M, Mirapeix RM, Castañer E, Domingo C, Mata JM, Marin A. Idiopathic azygos
14 vein aneurysm: a rare cause of mediastinal mass. *Thorax*. 1999;54:653–5.
- 15 7. Irurzun J, de España F, Arenas J, García-Sevila R, Gil S. Successful endovascular treatment
16 of a large idiopathic azygos arch aneurysm. *J Vasc Interv Radiol*. 2008;19:1251–4.
- 17 8. Savu C, Melinte A, Balescu I, Bacalbasa N. Azygos vein aneurysm mimicking a
18 mediastinal mass. *In Vivo*. 2020;34:2135–40.

1 9. Bobbio A, Miranda J, Gossot D, Perniceni T, Grunenwald D. Azygos vein aneurysm as a
2 diagnostic pitfall. The role of thoracoscopy. *Surg Endosc.* 2001;15:1049–50.

3 10. Takamori S, Oizumi H, Utsunomiya A, Suzuki J. Thoracoscopic removal of an azygos
4 vein aneurysm with thrombus formation. *Gen Thorac Cardiovasc Surg.* 2021;69:1335–7.

5

6

7

8

9

10

11

12

13

14

15

16

17

18

1 **Figure Legends**

2 **Fig.**

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

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

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

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

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

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

18 **(f, g)** Two streamline images of 4D-flow MRI in the venous system with different phases of the

1 cardiac cycle. A slightly slow turbulent flow was observed.

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

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

5

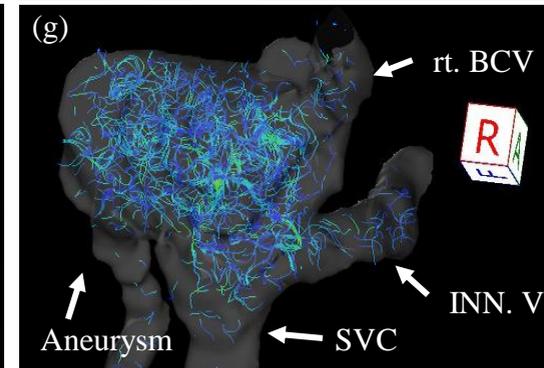
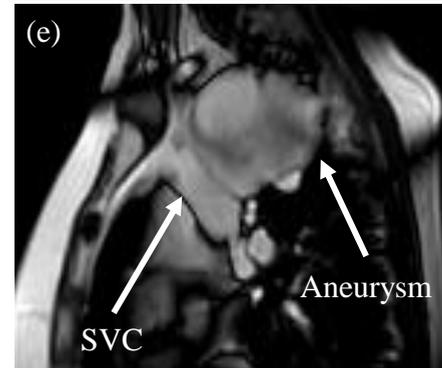
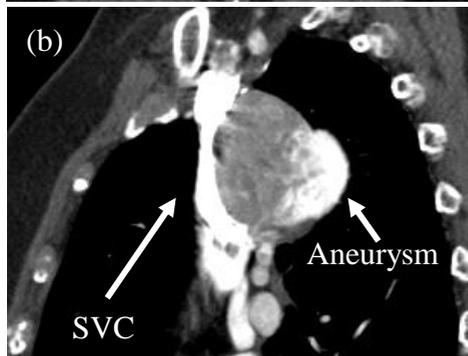
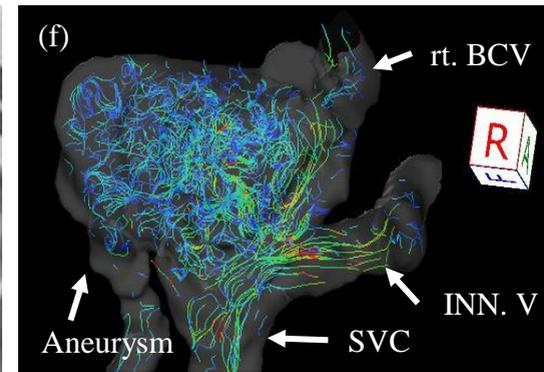
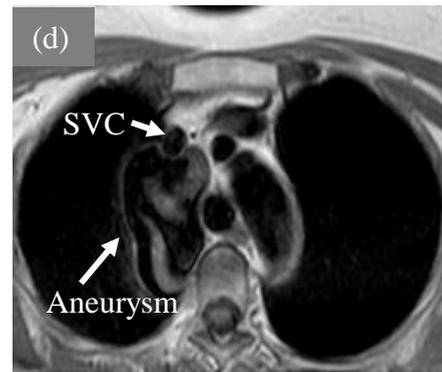
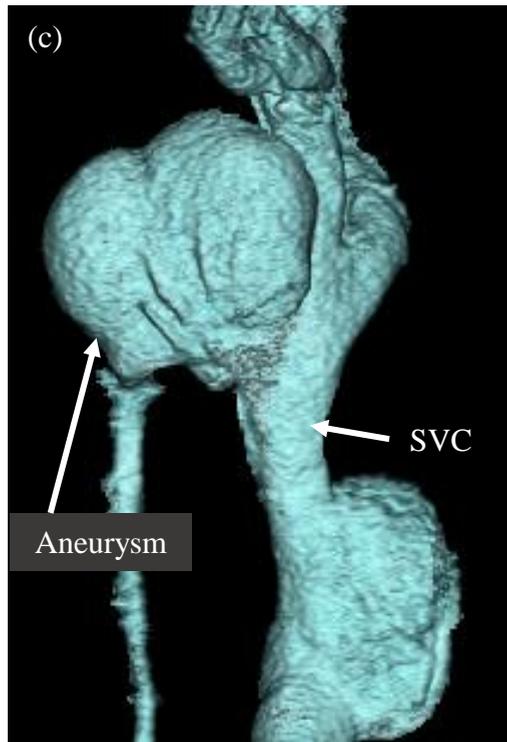
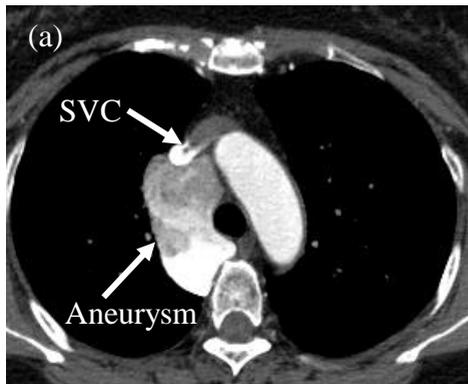
6 **Online Resource**

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

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

Dynamic CT

4D-Flow MRI



SVC : Superior vena cava rt. BCV : right Brachiocephalic Vein INN. V : Innominate Vein

Intraoperative thoracoscopic imaging.

