Radiology Case Reports 19 (2024) 3345–3348

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Exceptional multisite aneurysms in Takayasu arteritis: A unique and challenging case ☆,☆☆

Ermilo Echeverria-Ortegon[a,*], *Jose Luis Millet-Herrera*[a], *Ricardo Méndez-Molina*[a], *Javier Casillas*[b]

[a] *School of Medicine, Universidad Marista de Mérida, Periférico Norte Tablaje Catastral 13941, Temozón Nte, 97300 Mérida, Yuc*

[b] *Department of Radiology, University of Miami Health System, Miller School of Medicine, 1320 S Dixie Hwy, Coral Gables, FL 33146, EE. UU, Miami, FL, USA*

ARTICLE INFO

Article history:
Received 2 January 2024
Accepted 20 April 2024

Keywords:
Takayasu arteritis
Aneurysm

ABSTRACT

This case describes a rare and complex presentation of Takayasu arteritis, a large vessel vasculitis primarily affecting young females. Unlike typical Takayasu arteritis cases characterized by arterial stenosis, this 34-year-old male presented with an unusually high number of aneurysms affecting the aorta, subclavian arteries, and other segments. This unique abundance of aneurysms complicates diagnosis and management. This disease typically manifests as arterial stenosis, with aneurysms occurring in a minority of cases. The most common site for aneurysms is the ascending aorta, making multifocal aneurysms, as seen in this case, exceptionally rare. Managing multiple aneurysms in Takayasu arteritis is complex, necessitating careful consideration of factors like aneurysm size, morphology, and risk of complications. This case underscores the unique challenges posed by multifocal aneurysms in this condition, highlighting the need for a comprehensive approach to treatment.

Introduction

Takayasu arteritis (TAK) also known as “pulseless disease”, “aortic arch syndrome” is a large vessel vasculitis (inflammation of large blood vessels, such as the aorta and its main branches) that occurs more commonly in young females, generally in those less than 40 years of age. It was first described by Mikito Takayasu, an ophthalmologist at Kanazawa University in Japan, as a case of retinal vasculitis with pulselessness in 1908. This large vessel vasculitis is a rare autoimmune, idiopathic, granulomatous chronic disease, characterized by stenosis or occlusion of the aorta and its main branches, especially of the subclavian, common, and internal carotid arteries due to a variety of nonspecific inflammatory (systemic and local) and ischemic symptoms which leads to thickening of the arterial wall [1,2].

☆ Acknowledgments: None.

☆☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author.
E-mail address: milocho2000@gmail.com (E. Echeverria-Ortegon).

https://doi.org/10.1016/j.radcr.2024.04.056

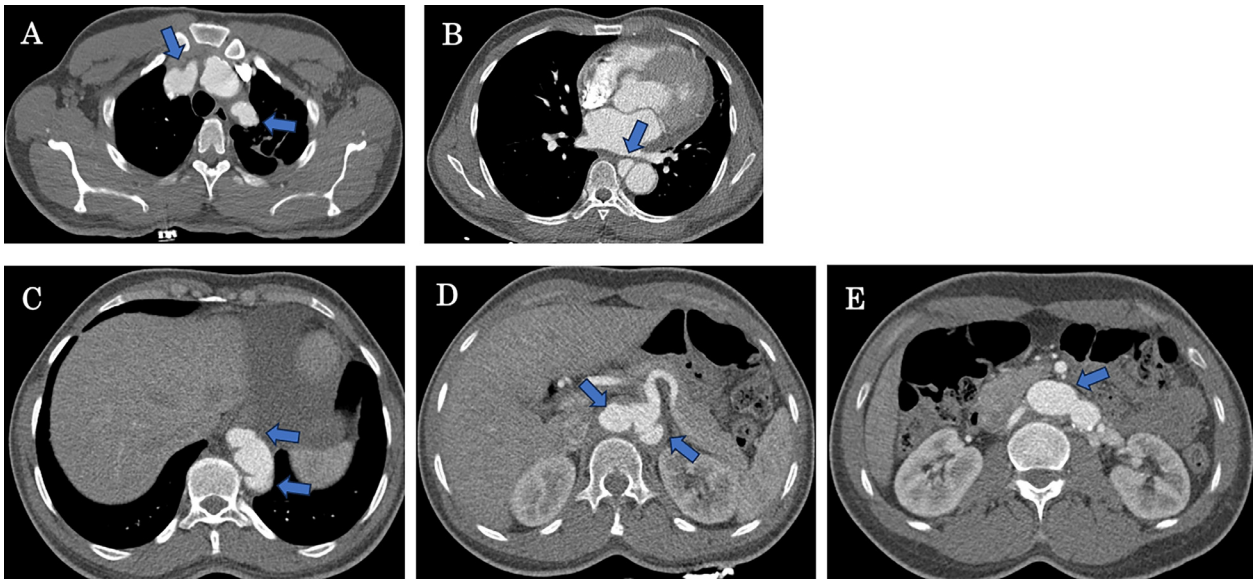

Fig. 1 – Contrast enhanced thoracic and abdominal computed tomography axial images (A-E) demonstrated multiple saccular aneurysms involving the thoracic, abdominal aorta and subclavian arteries (arrows).

Infrequently, the destruction of the elastica and muscularis layer may result in artery dilation and aneurysm, where most aneurysms occur in the ascending aorta and the subclavian arteries, and multiple aneurysmal involvements of the subclavian axillary, abdominal aortoiliac, lower extremity, and coronary arteries in TAK are extremely rare [3]. The aim of this case report is to describe the clinical importance and management of multiple aneurysms in TAK.

Case report

A 34-year-old male, with a history of Takayasu disease and thoracic and abdominal aorta aneurysms, who had been followed by Rheumatology Services in the past and history of cardiac catheterization that showed severe aortic insufficiency with cardiomyopathy with an ejection fraction of 25% presented to the emergency room with intermittent chest discomfort and associated shortness of breath and hemoptysis. Patient was under consideration for aortic valve replacement.

A thoracic and abdominal computed tomography study done at admission, showed the presence of multiple saccular aneurysms involving the thoracic and abdominal aorta and both subclavian arteries (Figs. 1A-E) (Figs. 2A-B). Patient was under consideration for aortic valve replacement.

Discussion

TAK is a large vessel vasculitis known for its propensity to affect women from childhood until the age of forty. TAK is rare, with an annual incidence of 1.2-2.6 cases/1 million. Most series reveal that 85% of cases are women, although the gender association has been shown to vary with geographic location [4]. The distribution of the disease is more common in Japan and the Indian subcontinent, but studies have shown an increasing incidence in Mexico, Brazil, and Colombia [3].

Typically, TAK presents with stenosis or occlusion rather than aneurysmal disease. The core feature of this disease is arterial inflammation, which can be variably associated with a systemic acute-phase response. Inflammatory lesions are characterized by arterial wall thickening, frequently resulting in remodeling of the arterial lumen due to myofibroblast proliferation [5].

In most series, 90% of patients suffer arterial stenoses, and only up to 25% aneurysmal disease. The most common site of aneurysm formation in patients with TAK is the aorta, followed by the subclavian artery, the brachiocephalic artery, and the common carotid artery [6]. Specifically, in the aorta the aneurysms most frequently occur in the ascending aorta, followed by either the thoracic or abdominal aorta [7,8].

Isolated aortic aneurysms have been reported frequently. However, cases of multiple aneurysm presentation are relatively rare. Jain et al described a case of a 20-year-old patient with the presence of multiple saccular aortic aneurysms in both the descending thoracic and abdominal aorta [9]. Studies by Matsumura et al and Yang et al found that while the incidence of saccular or fusiform aneurysms was 31%, the presence of multiple aneurysms was observed in only 13% and 12% of patients, respectively, making this a highly uncommon presentation [7,8].

We reported an infrequent case of a patient with known TAK disease, that presented multifocal aneurysmal dilatation involving the thoracic and upper abdominal aorta, as well as aortic arch branches. We observed an exceptionally high number of aneurysms, with a total of 12, in this patient with TAK, which stands out significantly when compared to the other clinical cases. This high quantity of aneurysms presents

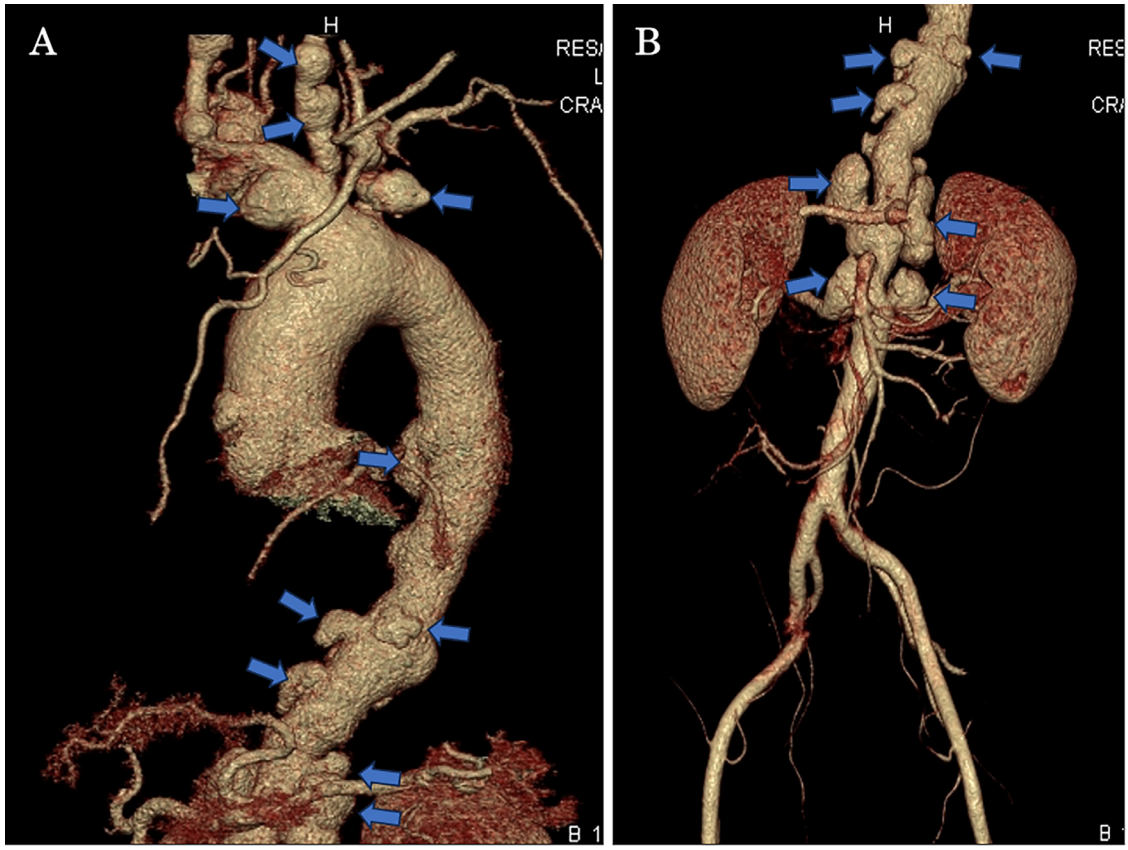

Fig. 2 – Volume rendering reconstructions coronal plane (A and B). Redemonstrated the presence of multiple saccular aneurysms involving the thoracic aorta, abdominal aorta, and subclavian arteries (arrows).

a formidable challenge in terms of treatment planning and management, particularly given the heightened risk of complications.

Despite the slow development of aortic aneurysm (with a reported mean growth rate of 0.04 cm/year), most patients are young with a longer life expectancy, and the risk of rupture poses a threat to the patient all the time, like the one we reported, as well as the previously reported cases. Additionally, there are other potentially fatal consequences, such as aortic regurgitation or heart failure [8]. Nonetheless, the lack of knowledge about multiple aneurysms risk in TAK patients due to the rarity of the disease indicates that the management strategies for aortic aneurysm in TAK should consider several factors, including the location and size of aneurysm, complexity of vessel lesions, and disease activity [10].

Generally, identification of the aneurysms in the aorta is indication for surgical intervention, so as to leave as little of the lesion as possible in order to avoid the occurrence of postoperative anastomotic dehiscence or pseudoaneurysm [11]. Kieffer et al reported satisfactory surgical outcome of descending thoracic and thoracoabdominal aortic isolated aneurysm in 33 patients with TAK where except in special cases (e.g., endoaneurysmorrhaphy associated with bypass and endovascular aortic repair), the standard technique for repair of descending thoracic or thoracoabdominal aortic aneurysm was prosthetic endovascular graft replacement [12]. Stent-graft implantation is an interesting therapeutic alternative to conventional open surgery for the treatment of the thoracic and abdominal aorta; it avoids replacement of the aorta, which has greater mortality. Moreover, endovascular repair is shorter and less invasive and has a lower risk of systemic complications.

However, in situations where TAK present with multiple aneurysms, a specific treatment approach is not well-defined, especially with a large number of aneurysms, like in our case. Single-stage surgery is generally preferable, especially for extensive thoracic aortic aneurysms, but due to the invasiveness of this method, there are instances where staged surgery becomes necessary. Therefore, when dealing with multiple aneurysms in TAK, the decision regarding the order of surgical treatment is determined based on factors such as the diameter, morphology, tendency for lesion expansion, and concerns about mortality risk between surgeries due to the potential of aneurysm rupture [13].

Patient consent

A written informed consent was obtained from the patient for the publication of this case report.

Data statement

The data of the case is available via inquiries from the corresponding author.

REFERENCES

[1] Keser G, Aksu K, Direskeneli H. Takayasu arteritis: an update. Turk J Med Sci 2018;48(4):681–97. doi:10.3906/sag-1804-136.

[2] Podgorska D, Podgorski R, Aebisher D, Dabrowski P. Takayasu arteritis - epidemiology, pathogenesis, diagnosis and treatment. J Appl Biomed 2019;17(1):20 Epub January 9, 2019. doi:10.32725/jab.2018.005.

[3] Wang J, Li C, Zheng Y, Liu J, Sheng S, Yuan Z, et al. Multiple aneurysms of the subclavian-axillary, abdominal aortoiliac, lower extremity, and coronary arteries in a pediatric patient of Takayasu arteritis. Ann Vasc Surg 2022;81:387.e1 Epub October 15, 2021. doi:10.1016/j.avsg.2021.08.012.

[4] Wetstein PJ, Clark ME, Cafasso DE, Golarz SR, Ayubi FS, Kellicut DC. Surgical repair of abdominal aortic and renal artery aneurysms in Takayasu's arteritis. Hawaii J Med Public Health 2016;75(1):4–7.

[5] Tombetti E, Mason JC. Takayasu arteritis: advanced understanding is leading to new horizons. Rheumatology (Oxford) 2019;58(2):206–19. doi:10.1093/rheumatology/key040.

[6] Oura K, Taguchi K, Yamaguchi Oura M, Itabashi R, Maeda T. Takayasu's arteritis with a thrombosed aneurysm on the common carotid artery causing ischemic stroke. Intern Med 2022;61(3):425–8 Epub July 30, 2021. doi:10.2169/internalmedicine.7735-21.

[7] Matsumura K, Hirano T, Takeda K, Matsuda A, Nakagawa T, Yamaguchi N, et al. Incidence of aneurysms in Takayasu's arteritis. Angiology 1991;42(4):308–15 PMID: 1673052. doi:10.1177/000331979104200408.

[8] Yang KQ, Meng X, Zhang Y, Fan P, Wang LP, Zhang HM, et al. Aortic aneurysm in Takayasu arteritis. Am J Med Sci 2017;354(6):539–47 Epub September 1, 2017. doi:10.1016/j.amjms.2017.08.018.

[9] Jain, Neeraj & Chauhan, Udit & Jain, Shweta & Goel, Vandana & Puri, Sunil. (2018). Multiple aneurysms in Takayasu arteritis: an uncommon presentation Clinical History. 10.1594/EURORAD/CASE.12923.

[10] Pan L, Du J, Chen D, Zhao Y, Guo X, Qi G, et al. Takayasu arteritis with dyslipidemia increases risk of aneurysm. Sci Rep 2019;9(1):14083. doi:10.1038/s41598-019-50527-z.

[11] Iida Y, Fujii S, Sawa S, Shimizu H. Successful treatment of giant ascending aortic aneurysm with Takayasu arteritis. Int Heart J 2020;61(2):397–9 Epub March 14, 2020. doi:10.1536/ihj.19-355.

[12] Kieffer E, Chiche L, Bertal A, Koskas F, Bahnini A, Blã Try O, et al. Descending thoracic and thoracoabdominal aortic aneurysm in patients with Takayasu's disease. Ann Vasc Surg 2004;18(5):505–13 Epub August 6, 2004. doi:10.1007/s10016-004-0073-y.

[13] Regina G, Bortone A, Impedovo G, De Cillis E, Angiletta D, Marotta V. Endovascular repair of thoracic stent-graft bulging rupture in a patient with multiple thoracic aneurysms due to Takayasu disease. J Vasc Surg 2007;45(2):391–4. doi:10.1016/j.jvs.2006.10.001.