



Native bicuspid aortic valve thrombus in a patient with an ascending aorta aneurysm: A case report

Shuofang Ren ^{*,1}, Yulin Wen, Guotao Ma, qi Miao

Department of Cardiac Surgery, Peking Union Medical College Hospital, Peking Union Medical College, Chinese Academy of Medical Science, Beijing, China

ARTICLE INFO

Keywords:

Thrombus formation
Bicuspid aortic valve
Ascending aorta aneurysm
Coronary angiography
Case report

ABSTRACT

Thrombus formation on a well-conserved bicuspid aortic valve is rare. We encountered a patient with organized thrombus formation on a native bicuspid aortic valve without calcification or stenosis, which was found occasionally during an elective operation for ascending aorta replacement surgery. The location of the thrombus was just at the orifice of left coronary artery, which produced the atherosclerosis-like symptoms such like exertional chest tightness and dyspnea. And these are no apparent predisposing causes of thrombosis could be ascertained postoperatively. The patient is in excellent condition 6 months after the operation. The lesson we learned from our case is that when the patient's symptom can't correspond with his or her diagnosis, we should ask more questions, evaluate the patient thoroughly and make the differential diagnosis as possible as we can. And the surgery can be performed aggressively when patient's symptoms cannot be figured out by physical examination, not only for pathologic confirmation but also for the prevention of life-threatening complications that can caused by either condition.

1. Introduction

We present the case of a patient with an organized thrombus formation on a native bicuspid aortic valve (BAV), without calcification or stenosis, found incidentally during elective ascending aorta replacement surgery. No apparent predisposing causes of thrombosis were identified postoperatively. The patient was in excellent condition 6 months postoperatively.

1.1. Case presentation

A 58-year-old male, with an unremarkable medical history, presented to our institution with a 2-year history of chest tightness on exertion and shortness of breath (SOB). He had two episodes of acute onset syncope with an unknown cause. Multidetector computed tomography angiography revealed an ascending aortic aneurysm (Fig. 1A), the diameters of the ascending aorta and sinus of Valsalva were 49 and 40 mm, respectively. Transthoracic echocardiography revealed mild aortic regurgitation of the BAV. Since the ascending aortic aneurysm and mild aortic regurgitation could not explain the patient's symptoms, which included chest tightness, dyspnea, and

* Corresponding author. Peking Union Medical College Hospital (Dongdan campus), No.1 Shuaifuyuan Wangfujing Dongcheng District, Beijing, 100730, China.

E-mail address: miaoqipumc@hotmail.com (S. Ren).

¹ First author.

<https://doi.org/10.1016/j.heliyon.2023.e18463>

Received 24 December 2022; Received in revised form 13 July 2023; Accepted 18 July 2023

Available online 20 July 2023

2405-8440/© 2023 Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

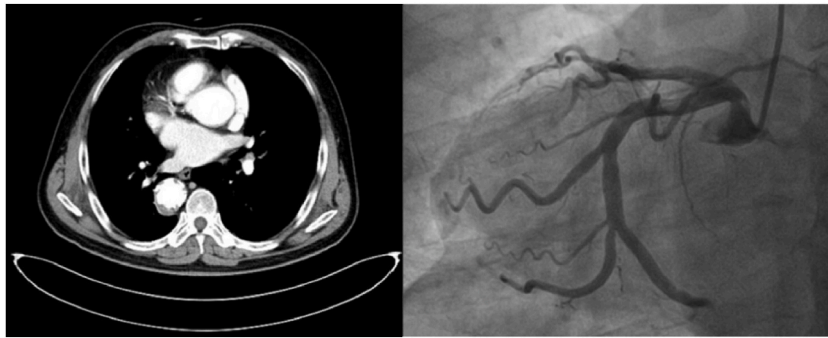


Fig. 1. (A) Multidetector computed tomography angiography revealed an ascending aortic aneurysm. (B) Coronary angiography showed that blood perfusion was quite slow in the left coronary artery but with no evidence of significant atherosclerosis (supplementary files can see this process obviously).

syncope, other probable causes were explored. Atherosclerosis could generate exertional chest tightness and SOB, while cerebrovascular disease and Stokes-Adams might trigger syncope. Therefore, coronary angiography, transcranial Doppler (TCD), and ambulatory electrocardiography monitoring were performed. Coronary angiography demonstrated slow blood perfusion in the left coronary artery (LCA), but there was no evidence of significant atherosclerosis (Fig. 1B and Supplementary Video). No abnormalities were seen on TCD and a Holter monitor. According to the American College of Cardiology/American Heart Association valve disease guidelines, patients with BAVs, with smaller aortic diameters, should undergo surgery. Thus, an elective ascending aortic replacement surgery was performed.

After median sternotomy and initiation of cardiopulmonary bypass, the ascending aorta was cross-clamped and incised during cardiac arrest. After aortotomy, the BAV with left-right fusion was present. An incidental, organized thrombus was exposed at the LCC of the aortic valve, directly covering most of the LCA's ostium (Fig. 2A and B). However, the mass attached to the aortic surface of the LCA cusp was not seen on transesophageal echocardiography. The thrombus measured approximately 1×0.8 cm. After excision of the thrombus, inspection of the valve showed no abnormalities. Thus, valve replacement was not necessary. Histopathological examination confirmed that the excised mass was an organized thrombus (Fig. 3). Thrombus formation on the orifice of the LCA explains the patient's exertional chest tightness, SOB, and syncope, and was likely to cause the changes seen on left coronary angiography. The patient's postoperative course was uncomplicated.

Thrombus formation is associated with a hypercoagulable state, certain autoimmune diseases, and malignancies. His postoperative blood examination included antithrombin III, protein C, protein S, C3/C4, fibrinogen, prothrombin time, plasminogen, blood count, erythrocyte sedimentation rate, lupus anticoagulants, anticardiolipin antibodies, antiphospholipid antibodies, and tumor markers. Microbiological tests, including blood and tissue cultures were negative, and there was no evidence of infective endocarditis. The patient was discharged on the fifth postoperative day. A 3-month therapy of warfarin and lifelong aspirin were prescribed as anticoagulants.

2. Discussion

We present a case of sinus Valsalva thrombosis in a patient with an ascending aneurysm and BAV. The thrombus was located at the orifice of the LCA, and produced atherosclerosis-like symptoms such as exertional chest tightness and dyspnea. We attempted to ascertain the primary cause of native aortic valve thrombosis by performing blood tests screening for coagulation disorders,

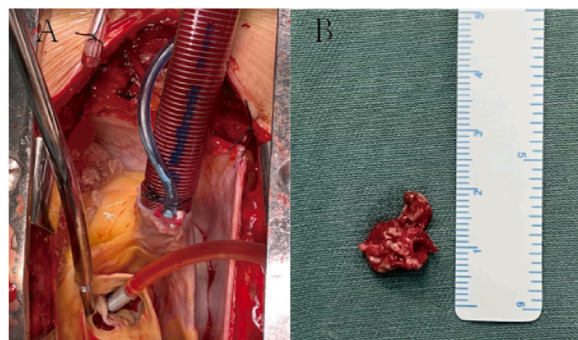


Fig. 2. (A) Mass was exposed at the left coronary cusp of aortic valve just after aortotomy. The intraoperative photograph demonstrates that the surface of the aortic valve seemed to be normal and the leaflets were thin. (B) Macroscopic image of 1.0×0.8 cm sized thrombotic mass.

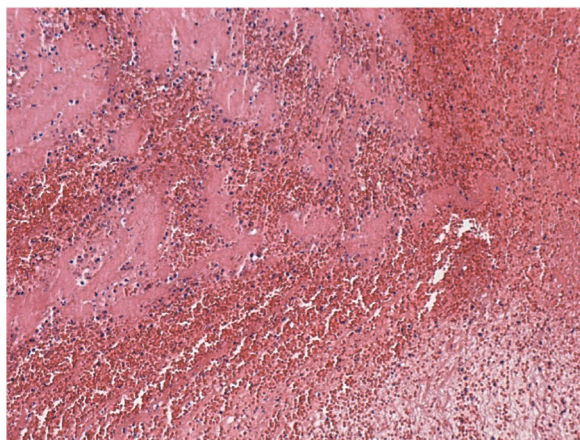


Fig. 3. Excised mass was confirmed as an organized thrombus by microscopic study.

autoimmune diseases, and tumor markers, all of which were negative. He had unremarkable results, except for bicuspid malformation and an ascending aortic aneurysm. Organized thrombus formation in the native aortic valve has been reported as a rare complication of BAV [1]. Thrombi with severe calcification or stenosis is commonly observed on the valve surface with severe calcification or stenosis [2,3]. However, in our case, the aortic valve was well-conserved and functioning, and the organic lesion in the AV was mild. Toshimi reported a case of an aortic bicuspid valve with thrombus formation without severe stenosis and calcification. However, this patient's condition was complicated by moderate aortic regurgitation, which caused a swirling current on the LV side, and an organized thrombus was directly present on the valve surface [4]. We hypothesized that BAV may be a risk factor for thrombus formation.

BAV is the most common congenital heart malformation in 1–2% of the general population [5]. It carries a higher risk of complications such as aortic dilation, valvular dysfunction, and even aortic valve thrombosis, which are commonly related to coagulation disorders and may even occur spontaneously [2]. When two of the three leaflets of the AV fuse, the asymmetric BAV morphology alters the normal application of the downstream jet and flow structures or patterns, and the swirling current produces shear stress that injures the valve [6,7]. In addition, both the Valsalva sinus asymmetry and the interaction between the raphe and commissure may lead to a tilted deployment that promotes blood stasis in these areas. Therefore, hemodynamic changes caused by morphological abnormalities in the BAV may increase the propensity for microthrombus formation.

3. Conclusion

This case highlights that in patients presenting with exertional chest tightness and syncope, that can't be explained by an ascending aneurysm and BAV, a thorough evaluation should be performed. Echocardiography can be performed to focus not only on the cardiac cavity but also on the AV surface and sinus of Valsalva, to determine any structural abnormalities. Spontaneous aortic thrombosis in patients with BAV is a rare condition that can cause catastrophic events. One case reported by Han revealed an acute ST-elevation myocardial infarction caused by idiopathic native aortic valve thrombosis in a healthy middle-aged man with no established cardiovascular risk factor [8]. In addition, systemic thromboembolism, including cerebrovascular events, may occur secondary to embolization from the aortic thrombosis of the BAV. Decision-making based on risk/benefit analysis may be challenging. Therefore, surgery may be required when the patient's symptoms cannot be determined by physical examination, not only for pathologic confirmation, but also for preventing life-threatening complications that can be caused by either condition.

Ethics statement

The authors confirm that written consent for the submission and publication of this case report, including images and associated text, was obtained from the patient in accordance with COPE guidelines.

Funding

None declared.

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Data availability statement

Data will be made available on request.

Additional information

No additional information is available for this paper.

Declaration of competing interest

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.

Acknowledgments

None declared.

References

- [1] S.M. Yuan, H. Sm, H. Jing, The bicuspid aortic valve and related disorders, *Sao Paulo Med. J.* 128 (5) (2010) 296–301, <https://doi.org/10.1590/s1516-31802010000500010>. PMID: 21181071.
- [2] N.V. Mahajan, Khetarpal, V. L, L. Afonso, Stroke secondary to calcific bicuspid aortic valve: case report and literature review, *J. Cardiol.* 54 (1) (2009) 158–161, <https://doi.org/10.1016/j.jjcc.2008.10.011>. Epub 2008 Dec 24. PMID: 19632538.
- [3] S. Wan, , et al.S, J.M. DeSmet, J.L. Vincent, J.L. LeClerc, Thrombus formation on a calcific and severely stenotic bicuspid aortic valve, *Ann. Thorac. Surg.* 64 (2) (1997) 535–536, [https://doi.org/10.1016/S0003-4975\(97\)00545-6](https://doi.org/10.1016/S0003-4975(97)00545-6). PMID: 9262609.
- [4] T. Koitabashi, , et al.T. Inomata, T. Kaida, H. Shinagawa, K. Shibata, K. Miyaji, et al., A case of aortic bicuspid valve with thrombus formation without severe stenosis and calcification, *J. Echocardiogr.* 10 (4) (2012) 148–150, <https://doi.org/10.1007/s12574-012-0147-8>. Epub 2012 Oct 6. PMID: 27278354.
- [5] I. Mordi, N, N. Tzemos, Bicuspid aortic valve disease: a comprehensive review, *Cardiol. Res. Pract.* 2012 (2012), 196037, <https://doi.org/10.1155/2012/196037>. Epub 2012 May 28. PMID: 22685681; PMCID: PMC3368178.
- [6] A. Pasipoularides, Historical continuity in the methodology of modern medical science: leonardo leads the way, *Int. J. Cardiol.* 171 (2) (2014) 103–115, <https://doi.org/10.1016/j.ijcard.2013.11.133>. Epub 2013 Dec 7. PMID: 24360160.
- [7] A. Pasipoularides, Calcific aortic valve disease: Part 1—Molecular pathogenetic aspects, hemodynamics, and adaptive feedbacks, *J. Cardiovasc. Transl. Res.* 9 (2) (2016) 102–118, <https://doi.org/10.1007/s12265-016-9679-z>. Epub 2016 Feb 18. PMID: 26891845; PMCID: PMC4833551.
- [8] D.C. Han, Dc, J.S. Kim, S.K. Lee, K.S. Choo, K.U. Choi, K.J. Chun, et al., Native aortic valve thrombosis: an unusual cause of acute ST-elevation myocardial infarction, *Cardiovasc. Pathol.* 22 (3) (2013) e23–e26, <https://doi.org/10.1016/j.carpath.2013.01.001>. Epub 2013 Jan 30. PMID: 23375581.