

CASE REPORT

DOI: 10.5336/caserep.2022-90451

Circumflex-to-Bronchial Artery Fistulas in Behçet's Disease

¹Duygu İMRE YETKİN^a, ²Selçuk AKKAYA^b, ³Tuncay HAZIROLAN^c

^aClinic of Radiology, Midyat State of Hospital, Mardin, Türkiye

^bDepartment of Radiology, Karadeniz Technical University Faculty of Medicine, Trabzon, Türkiye

^cDepartment of Radiology, Hacettepe University Faculty of Medicine, Ankara, Türkiye

ABSTRACT A coronary-to-bronchial artery fistula is a rare entity that emerged in the case of chronic pulmonary thromboembolism (cPTE). We present a case of a 50-year-old man with coronary-to-bronchial artery fistula in Behçet's disease that had been hospitalized in consequence of pulmonary thromboembolism attacks and treated with anticoagulation therapy repeatedly. After myocardial infarction was ruled out, coronary computerized tomography angiography was performed, and demonstrated right pulmonary artery occlusion, delay of the contrast passage in the right pulmonary veins, dilated right lung bronchial arteries, and fistulas between the right lung bronchial arteries and the left circumflex artery due to cPTE. Diagnosis of coronary-to bronchial artery fistula and early treatment are very important for the prevention of fatal complications. Radiologist and clinician should be alerted for this entity.

Keywords: Behçet's disease; bronchial artery; coronary artery; fistula

A coronary-to-bronchial artery fistula is a rare entity that emerged in the case of chronic pulmonary thromboembolism (cPTE). If a plan of embolization exists, it is crucial to demonstrate coronary-to-bronchial artery fistulas radiologically to avoid fatal consequences such as myocardial infarction. We present a case of coronary artery-to-bronchial artery fistula in Behçet's disease. Informed consent was given.

CASE REPORT

A 50-year-old man with Behçet's disease was admitted to our hospital for the evaluation of unstable angina pectoris. Blood test results and electrocardiogram ruled out myocardial infarction. Our patient had been suffering from Behçet's disease for 20 years. Initially, he had oral and genital painful, ulcerative lesions. For few times, he was treated with steroid therapy for panuveitis and arthritis. Pathergy test was positive. He had no neurologic or gastrointestinal involvement.

Imaging findings: Flash mode prospective electrocardiography-gated coronary computerized tomography (CT) angiography was performed with a dual-source CT system (Siemens Healthineers-SOMATOM Force, Erlangen, Germany), to show whether coronary artery pathology was the cause of angina pectoris. We used 70 milliliter (mL) of iohexol 350 milligram/100 mL as the contrast agent with 80 mL of 0.9% NaCl (sodium chloride) [at first, 10 mL NaCl at 5 mL/second (sec), then 60 mL contrast agent at 5 mL/sec, 10 mL contrast/40 mL NaCl mixture at 4 mL/sec, 30 mL NaCl at 4 mL/sec]. Coronary CT angiography demonstrated right pulmonary artery occlusion, delay of the contrast passage in the right pulmonary veins, dilated bronchial arteries, and fistulas between the right lung bronchial arteries and the left circumflex artery (LCx) due to the cPTE (Figure 1, Figure 2, Figure 3, Figure 4).

Treatment: He had been hospitalized as a consequence of PTE attacks and treated with anticoagulation therapy repeatedly. Since the patient's

Correspondence: Duygu İMRE YETKİN

Clinic of Radiology, Midyat State of Hospital, Mardin, Türkiye

E-mail: duyguimre0831@gmail.com



Peer review under responsibility of Türkiye Klinikleri Journal of Case Reports.

Received: 10 Apr 2022

Accepted: 31 Aug 2022

Available online: 08 Sep 2022

2147-9291 / Copyright © 2022 by Türkiye Klinikleri. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

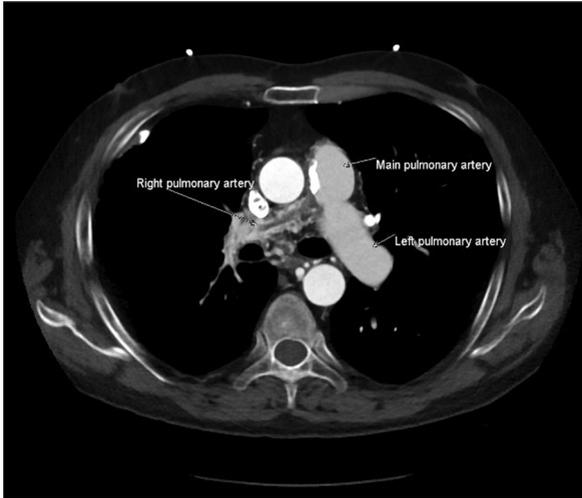


FIGURE 1: Axial coronary computerized tomography angiography shows occluded right pulmonary artery due to chronic thromboembolism in Behçet's disease. Also note dilated para-aortic bronchial arteries.

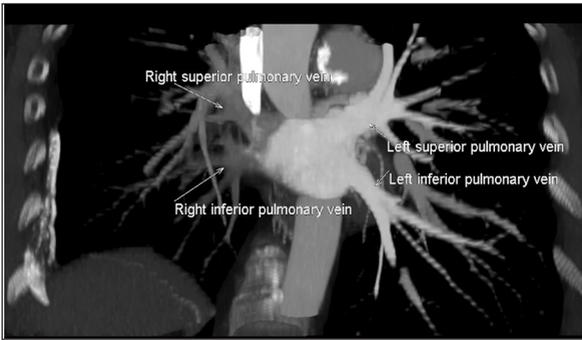


FIGURE 2: Coronal maximum intensity projection image shows delayed contrast passage in right pulmonary veins as a result of right pulmonary artery occlusion.



FIGURE 3: Axial oblique maximum intensity projection image shows tortuous and dilated bronchial artery-to-LCx fistulas (arrows). LCx: Left circumflex artery.

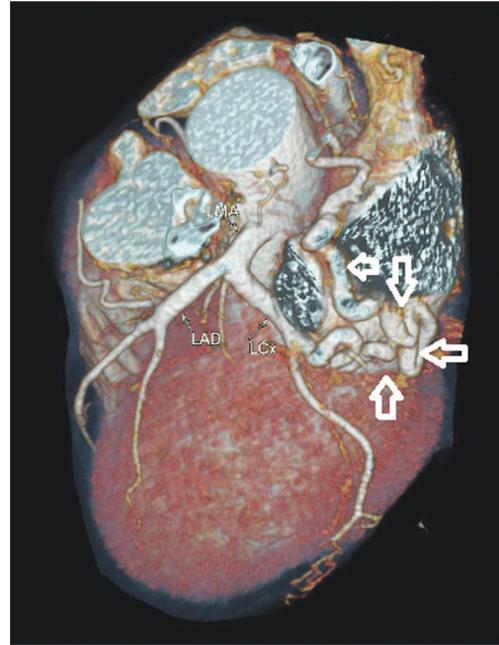


FIGURE 4: Volume-rendered image of coronary computerized tomography angiography shows dilated and tortuous LCx below left atrial appendage (hollow arrows). LMA: Left main coronary artery; LCx: Left circumflex artery; LAD: Left anterior descending artery.

international normalized ratio value was between 2-3, he was discharged with 5 mg warfarin per 24 hours, which he continued. Additional imaging was not performed because the patient did not come for a follow-up.

DISCUSSION

Behçet's disease is a multisystemic, chronic recurrent inflammatory disease with non-specific vasculitis involving vessels in several organs and systems in various degrees.¹ Oral and genital ulcerative lesions, skin lesions like erythema nodosum, positive pathergy test, arthritis, eye lesions like uveitis, thoracic, neurologic, gastrointestinal, vascular involvement are characteristic features of Behçet's disease.² The pulmonary artery is the second most frequent vascular structure involved after aorta.¹ The hypercoagulability in Behçet's disease is the underlying reason for the PTE.¹ The highest prevalence rate of Behçet's disease is in Türkiye with 80-370/100,000.² Behçet's disease may present with pulmonary stenosis or occlusion due to the recurrent PTE or with pulmonary artery aneurysms.¹ cPTE leads to dilatation of the

bronchial arteries.³ Fistulas between the coronary and bronchial arteries are rarely encountered and usually originated from the LCx.^{4,5} Coronary-to-bronchial artery fistulas may occur in chronic lung diseases, vasculitis such as Takayasu, and surgical interventions, and congenitally.⁶⁻⁸

Compensatory dilatation of bronchial arteries and fistulas between bronchial and LCx occurred in this case as a consequence of right pulmonary artery occlusion due to the cPTE as stated in the literature.¹

Coronary-to-bronchial artery fistula is a known entity.⁵⁻⁸ To our knowledge, no case of a fistula between the coronary and bronchial artery has been reported in Behçet's disease yet although coronary-to-bronchial artery fistulas are reported in other vasculitides like Takayasu.⁷ If a plan of embolisation exists, it is crucial to demonstrate coronary-to-bronchial artery fistulas radiologically to avoid fatal consequences such as myocardial infarction.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Duygu İmre Yetkin, Selçuk Akkaya, Tuncay Hazırolan; **Design:** Duygu İmre Yetkin, Selçuk Akkaya; **Control/Supervision:** Duygu İmre Yetkin, Selçuk Akkaya; **Data Collection and/or Processing:** Duygu İmre Yetkin; **Analysis and/or Interpretation:** Duygu İmre Yetkin, Selçuk Akkaya; **Literature Review:** Duygu İmre Yetkin; **Writing the Article:** Duygu İmre Yetkin; **Critical Review:** Duygu İmre Yetkin; **References and Fundings:** Duygu İmre Yetkin, Selçuk Akkaya; **Materials:** Duygu İmre Yetkin, Selçuk Akkaya.

REFERENCES

1. Chae EJ, Do KH, Seo JB, Park SH, Kang JW, Jang YM, et al. Radiologic and clinical findings of Behçet disease: comprehensive review of multi-systemic involvement. *Radiographics*. 2008;28(5):e31. [[Crossref](#)] [[PubMed](#)]
2. Erkan F, Gül A, Tasali E. Pulmonary manifestations of Behçet's disease. *Thorax*. 2001;56(7):572-8. [[Crossref](#)] [[PubMed](#)] [[PMC](#)]
3. Hasegawa I, Boisselle PM, Hatabu H. Bronchial artery dilatation on MDCT scans of patients with acute pulmonary embolism: comparison with chronic or recurrent pulmonary embolism. *AJR Am J Roentgenol*. 2004;182(1):67-72. [[Crossref](#)] [[PubMed](#)]
4. Papadopoulos DP, Perakis A, Votreas V, Anagnostopoulou S. Bilateral fistulas: a rare cause of chest pain. Case report with literature review. *Hellenic J Cardiol*. 2008;49(2):111-3. [[PubMed](#)]
5. Lee ST, Kim SY, Hur G, Hwang YJ, Kim YH, Seo JW, et al. Coronary-to-bronchial artery fistula: demonstration by 64-multidetector computed tomography with retrospective electrocardiogram-gated reconstructions. *J Comput Assist Tomogr*. 2008;32(3):444-7. [[Crossref](#)] [[PubMed](#)]
6. Byun SS, Park JH, Kim JH, Sung YM, Kim YK, Kim EY, et al. Coronary CT findings of coronary to bronchial arterial communication in chronic pulmonary disease. *Int J Cardiovasc Imaging*. 2015;31 Suppl 1:69-75. [[Crossref](#)] [[PubMed](#)]
7. Moon MH, Kang JK, Song H. Acquired coronary-to-bronchial artery fistula after two valve surgeries. *Asian Cardiovasc Thorac Ann*. 2014;22(4):478-80. [[Crossref](#)] [[PubMed](#)]
8. Maffei E, Bolognesi M, Cademartiri F. Complex congenital fistula between coronary arteries, bronchial arteries, and pulmonary artery assessed with cardiac computed tomography. *Eur Heart J*. 2017;38(26):2079. [[Crossref](#)] [[PubMed](#)]