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CASE REPORT

A rare cause of multiple pseudoaneurysms of radial artery after coronary angiography: Behçet's disease

Koroner anjiyografi sonrası çoklu radiyal arter yalancı anevrizmalarının nadir bir nedeni: Behçet hastalığı

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Summary– Patients with Behçet's disease are at risk for iatrogenic aneurysms after interventions. Presently described is the case of a 55-year-old male with multiple pseudoaneurysms occurring in the late period after a coronary angiography procedure performed via the radial artery. There was no previous diagnosis of vasculitis. Behçet's disease was revealed to be the underlying pathology.

Özet– Behçet hastaları girişimsel işlemler sonrasında iyatrojenik anevrizma gelişme riski taşımaktadırlar. Biz bu yazıda radiyal arter yoluyla yapılan koroner anjiyografi sonrası geç dönemde çoklu yalancı anevrizmalarla başvuran 55 yaşındaki erkek hastamızı sunmak istedik. Yaptığımız değerlendirme sonuçları, daha önce bilinen vaskülit tanısı olmayan hastanın Behçet tanısı almasını sağlamıştır.

Behçet's disease is a rare form of chronic vasculitis characterized by gastrointestinal, musculoskeletal, and neurological system involvement.[1] The disease usually appears in the third decade of life. Vascular involvement can be seen in approximately one-third of patients and in all sizes and types of vessels. Vascular involvement usually occurs within the first decade after the diagnosis. Venous involvement (6–25%) is more frequent than arterial involvement (1–5%) and usually occurs as superficial lower extremity thrombophlebitis or deep vein thrombosis. Arterial involvement may present with an aneurysm, or less frequently, stenosis or occlusion.[2,3] An aneurysm may occur in the aorta, pulmonary artery, or peripheral arteries. A pseudoaneurysm of the arteries is a rare manifestation of vascular involvement. During the active vasculitis stage of the disease, an intense infiltration of acute inflammatory cells occurs in the media and adventitia layers of the vessel. At this stage, the intima is swollen and the lumen is fully thrombosed. Thrombosis and occlusions usually occur in small arteries. In large vessels, the lumen is not fully thrombosed. When the vessel is not thrombosed and vasculitis has progressed, severe inflammation weakens the arterial wall and results in the formation of a pseudoaneurysm.[4] Rupture of a pseudoaneurysm has a high mortality rate.

It has been established that pseudoaneurysm formation may occur after trauma, especially following a vascular intervention. An intervention to the vessel wall or the insertion of a needle or catheter may induce either thrombosis or pseudoaneurysm formation at the puncture site. It has been reported that 17% to 50% of pseudoaneurysms occurred at puncture sites.[5,6]

In this report, the case of a patient with Behcet's disease and iatrogenic multiple pseudoaneurysms developing after a radial intervention is described.

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CASE REPORT

A 55-year-old male patient who had undergone a coronary angiography via the right radial artery was referred to the clinic with the complaint of swelling of the right wrist. A coronary angiogram revealed normal coronary arteries. The patient indicated that the swelling appeared in the fourth month after the angiography procedure. A pulsatile mass was palpated on the distal anterior part of the forearm medial to the radial artery and the radial artery pulse was determined to be intact in the physical examination (Fig. 1a). An ultrasound of the right wrist revealed a thrombosed pseudoaneurysm of the radial artery measuring 6x8 mm. The patient was boarded to the cardiovascular surgeon and the operation was not considered for its size and thrombosis. Within the following 1 to 2 weeks, another site of swelling occurred in the dorsal face of the right hand and a physical examination revealed a pulsatile mass in this region. Doppler ultrasonography revealed an 8x7-mm pseudoaneurysm originating in the radial arch between first and second metacarpal bones on the dorsal surface of the hand (Fig. 1b and c). During this period, painful swelling in the left ankle and the front of the leg occurred, and according to the laboratory tests performed at the time, the sedimentation rate was 25 mm/hour and the C-reactive protein level was 16.8 mg/L. The medical history revealed that patient had reported frequent oral aphthosis. With these findings, the patient was consulted to the department of rheumatology with a preliminary diagnosis of vasculitis. Large vessel vasculitis and pulmonary artery involvement were eliminated based on the results of thoracic magnetic resonance angiography and whole-body positron emission tomography-computed tomography imaging. Peripheral blood sampling was positive for the HLA-B 51 gene in a polymerase chain reaction assay. The results of a pathergy test were positive. A diagnosis of Behçet's disease was established according to the International Criteria for Behçet's Disease (oral aphthosis: 2 points, erythema nodosum-like painful lesions on the ankle and front of the leg: 1 point, vascular pseudoaneurysms: 1 point, positive pathergy test: 1 point).[7] The patient received 0.5 mg/kg steroid and azathioprine treatment and was scheduled for clinical follow-up.

DISCUSSION

Behcet's disease is a multisystem disorder with characteristic pathological findings of recurrent orogenital ulcers, vascular disorders, and ocular and cutaneous lesions. Behcet's disease is divided into 3 subtypes: neuro-, entero-, and vasculo-Behcet's disease. The primary manifestations of vasculo-Behcet's disease are aneurysm formation, arterial occlusion, and venous occlusion. Arterial involvement is present in 2% to 8% of patients and is associated with a high mortality.[8] The disease may present with aneurysms, pseudoaneurysms, or arterial occlusions. The pathogenesis of aneurysms in Behcet's disease is thought to be the obliteration of the vasa vasorum by the inflammatory process, resulting in a termination of the nutrient flow to the aortic wall. The intimal layer is thickened by fibroblasts and smooth muscle cells, whereas the media and adventitia are disrupted. Destruction of the media seems to be responsible for the development of saccular aneurysmal dilatation. Arterial reconstructions are also prone to thrombosis and pseudoaneurysm formation, as seen in our patient. Any invasive methods applied to the arterial system may cause pseudoaneurysms. Surgical treatment may lead to anasto-

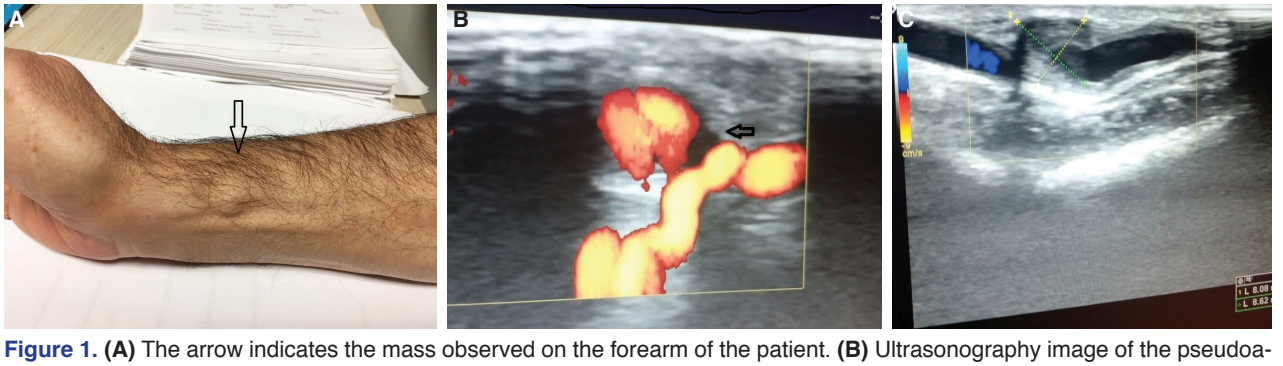


Figure 1. **(A)** The arrow indicates the mass observed on the forearm of the patient. **(B)** Ultrasonography image of the pseudoaneurysm of the radial artery. **(C)** Doppler image of the pseudoaneurysm of the radial artery.

motic false aneurysms. Surgical treatment should not be applied in the acute phase of the disease; systemic therapy, including colchicine, aspirin, and corticosteroids should be considered for all patients. Surgical treatment of pseudoaneurysms is problematic because new pseudoaneurysms frequently form at the site of the surgical graft or patch. An ultrasound-guided percutaneous thrombin injection was recently introduced as a technique to treat iatrogenic femoral pseudoaneurysms.[9,10] At present, there are no data addressing the indications for surgical intervention in the event of aneurysm formation. However, a patient with large pseudoaneurysms involving large vessels may be a candidate for surgical or percutaneous intervention due to the risk of a fatal pseudoaneurysm rupture.

In this case, pseudoaneurysms were observed after a coronary angiography using a transradial approach. Interestingly, the pseudoaneurysms were observed not only at the puncture site, but also other portions of the artery. This may have been the result of systemic inflammation induced by the radial artery puncture.

In this patient, we believe that the late pseudoaneurysm formation was associated with the radial intervention. Since the patient was asymptomatic before the intervention, and given that most pseudoaneurysms occur after interventions to the vessel wall, we thought that this radial pseudoaneurysm was associated with the radial intervention. Slow inflammation of the vessel wall may be the reason for the late presentation.

In conclusion, patients with Behcet's disease must be followed carefully after invasive procedures.

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REFERENCES

1. Jenkins AM, Macpherson AI, Nolan B, Housley E. Peripheral aneurysms in Behĉet's disease. Br J Surg 1976;63:199–202.
2. Gao N, Han W, Ci WP, Liao H, Du J. Clinical data analysis of cardiovascular involvement in Behcet's disease. [Article in Chinese] Zhonghua Yi Xue Za Zhi 2016;96:1523–6.
3. Seyahi E. Behçet's disease: How to diagnose and treat vascular involvement. Best Pract Res Clin Rheumatol 2016;30:279–95.
4. Cho SB, Cho S, Bang D. New insights in the clinical understanding of Behçet's disease. Yonsei Med J 2012;53:35–42.
5. Koç Y, Güllü I, Akpek G, Akpolat T, Kansu E, Kiraz S, et al. Vascular involvement in Behçet's disease. J Rheumatol 1992;19:402–10.
6. Lê Thi Huong D, Wechsler B, Papo T, Piette JC, Bletry O, Vitoux JM, et al. Arterial lesions in Behçet's disease. A study in 25 patients. J Rheumatol 1995;22:2103–13.
7. International Team for the Revision of the International Criteria for Behçet's Disease (ITR-ICBD). The International Criteria for Behçet's Disease (ICBD): a collaborative study of 27 countries on the sensitivity and specificity of the new criteria. J Eur Acad Dermatol Venereol 2014;28:338–47.
8. Calamia KT, Schirmer M, Melikoglu M. Major vessel involvement in Behçet's disease: an update. Curr Opin Rheumatol 2011;23:24–31.
9. Ozguler Y, Hatemi G, Yazici H. Management of Behcet's syndrome. Curr Opin Rheumatol 2014; 26:285–291.
10. Ding ZY, Jin GN, Ai X, Li LY, Zheng P, Guan Y, et al. Endovascular Treatment of Behcet Disease With Recurrent Infrainguinal Arterial Pseudoaneurysms: A Case Report and Literature Review. Medicine (Baltimore) 2016;95:e3545.

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Anahtar sözcükler: Behçet hastalığı; koroner anjiyografi; iyatrojenik; yalancı anevrizma; radiyal arter.