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Case Report / Olgu Sunumu

Pulmonary artery aneurysm leading to coronary ischemia

Koroner iskemiye neden olan pulmoner arter anevrizması

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Pulmonary artery aneurysm is a rare clinical entity and its definitive diagnosis requires a high degree of clinical suspicion. In this article, we report on a 27-year-old female patient admitted with the signs and symptoms of coronary ischemia due to the compression of pulmonary artery aneurysm on the left main coronary artery. The patient underwent successful surgical correction.

Key words: Chest pain; coronary ischemia; pulmonary artery aneurysm.

Pulmoner arter anevrizması nadir görülen klinik bir durum olup, kesin tanısı yüksek derecede klinik şüphe gerektirir. Bu yazıda, pulmoner arter anevrizmasının sol ana koroner artere basısı nedeniyle oluşan semptom ve bulgularla başvuran 27 yaşında bir kadın olgu sunuldu. Hasta başarılı bir cerrahi düzeltme geçirdi.

Anahtar sözcükler: Göğüs ağrısı; koroner iskemi; pulmoner arter anevrizması.

A pulmonary artery aneurysm (PAA) is a rare clinical entity that makes up less than 1% of all aneurysms in the thoracic cavity.[1,2] It can be either congenital or acquired,[3,4] and the symptom-free nature of the pathology of PAAs may lead to a huge expansion of the aneurysm before a definitive diagnosis can be established. However, patients may infrequently present with non-specific symptoms related to the compression of the surrounding vital tissues by the aneurysmal sac. Herein, we present a young female patient who had the signs and symptoms of coronary ischemia, but an evaluation revealed a secundum-type atrial septal defect (ASD) along with main and bilateral branch PAAs that were successfully treated with surgical intervention.

CASE REPORT

A 27-year-old female patient was referred to our clinic with chest pain and dyspnea upon exertion. On her physical examination, she was in sinus rhythm and normotensive. In addition, there were prominent pulsations and a grade 3/6 systolic murmur over the left sternal border. Chest radiography showed an enlarged pulmonary arterial shadow with moderate cardiomegaly, and tests for systemic and infectious diseases yielded negative results. Furthermore, ST-segment depression on anterior chest derivations was also noted while transthoracic echocardiography (TTE) revealed a secundum-type ASD (1.4 cm), large PAA, and mild-to-moderate pulmonary insufficiency. Subsequent computed tomography (CT) confirmed the initial diagnosis and demonstrated a main pulmonary aneurysm measuring 51 mm in length that involved both the right and left branches up to the hilar portions (Figure 1). Additionally, a tomographic examination demonstrated compression of the left main coronary artery (LMCA) by the aneurysmal sac at its proximal portion (Figure 2), and further examination via cardiac catheterization documented a mean pulmonary arterial pressure (mPAP) of 33 mmHg and a wedge pressure of 5 mmHg.

The operation was carried out through a median sternotomy. Following a pericardiotomy, aneurysmal dilatation of the main and branch pulmonary arteries was also noted. The first part of the procedure was performed with a beating heart on normothermic

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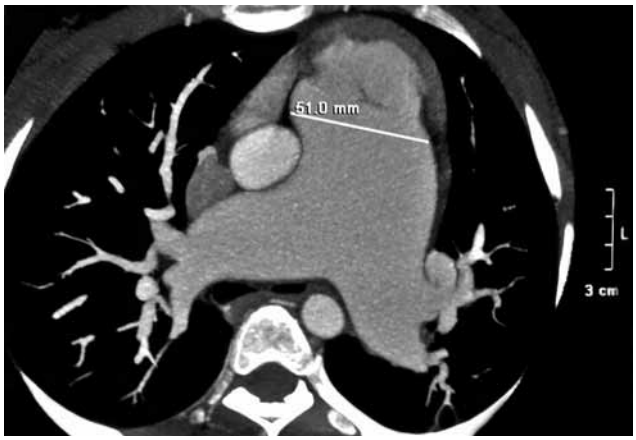

Figure 1. Computed tomography showed the main pulmonary aneurysm involving both the right and left branches up to the hilar portions.

cardiopulmonary bypass (CPB) with standard aortic and bicaval cannulations. The main, right, and left pulmonary arteries were then dissected free up to the hilar portions, and the enlarged, diseased portions of the pulmonary arteries were completely excised. A Dacron graft (28 mm) was then interposed between the hilar portions of the right and left pulmonary arteries using a running technique with 4-0 prolene sutures, and a 32 mm Dacron graft was then anastomosed end-to-side to the previously interposed graft. The rest of the operation was performed with a cross-clamp at normothermia. After cardiac arrest, a right atriotomy was executed, and the secundum-type defect was primarily repaired. The pulmonary valve was then inspected and found to be grossly normal and competent. Therefore, the pulmonary valve was preserved, and the proximal end of the 32 mm Dacron graft was sutured to the proximal aspect of the main pulmonary artery. Next, the patient was weaned off of CPB with minimal inotropic support. The postoperative course was uneventful, and she was extubated after six hours and discharged on the sixth postoperative day. A histopathological examination of the biopsy material taken from the arterial wall confirmed the preoperative diagnosis, but the results were negative for any systemic inflammatory or infectious diseases. Postoperative control echocardiography was satisfactory, showing only trivial pulmonary regurgitation, and there was no abnormality in the postoperative control CT (Figure 3).

DISCUSSION

Autopsies have revealed an estimated incidence rate for PAAs of one in 14,000,[5] but aneurysms involving both the main and branch pulmonary arteries are extremely rare. Hence, there are no established guidelines regarding the management of this clinical entity. In addition, the surgical indications are not clear, but an operation is sometimes advised in cases involving right ventricular dysfunction, severe valvular incompetence, and progressive aneurysmal dilatation or when common symptoms of PAAs such as dyspnea, cough, and chest pain develop.[3] However, these are

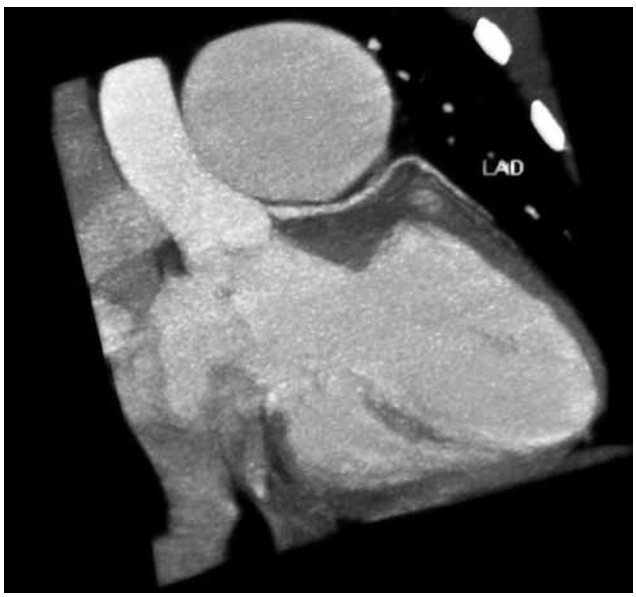

Figure 2. Compression of the left main coronary artery by the aneurysmal sac at its proximal portion.

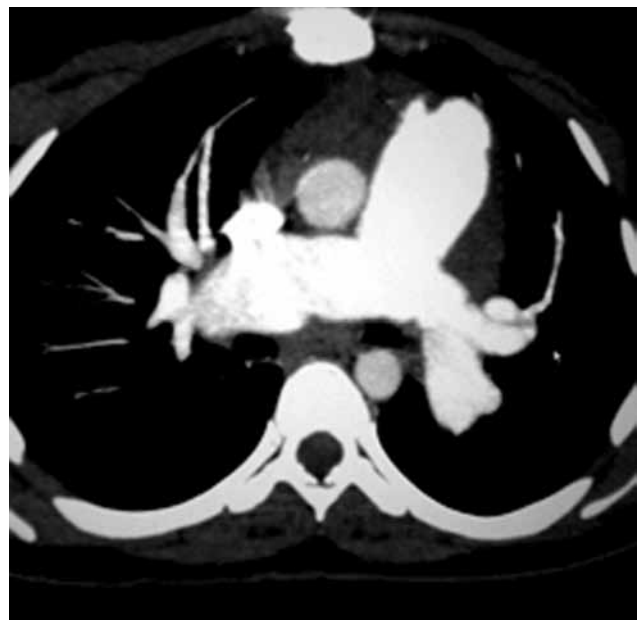

Figure 3. Postoperative control tomography of the patient.

generally non-specific, and the diagnosis requires a high degree of clinical suspicion, as was the case in our patient.

The most common pathologies associated with PAA are congenital cardiac anomalies, which constitute more than 50% of the etiologies, but other causes such as infections (endocarditis, syphilis, and tuberculosis), arteriosclerosis, cystic medial necrosis, vasculitis, hypertension (HT), trauma, and arteriovenous fistulas (AVFs) may also be involved.[2] Behçet's disease (BD), a multisystemic vasculitis of unknown etiology, is a well known disease in Turkey that causes arterial aneurysms, and PAAs can develop over the course of this systemic disease.[6,7] Therefore, BD and other possible etiologies were also considered in the differential diagnosis of our patient to avoid any misdiagnosis. Fortunately, the systemic work-up and histopathological examination of the biopsy material taken from the diseased arterial wall were completely negative for any other disease, and the ASD was accepted as the underlying cause of the PAA.

Since there is scanty data in the medical literature, a clear guideline has not been established for the treatment of PAAs, and the approach to surgical repair remains controversial, with various techniques that include aneurysmorrhaphy, wall plication, pericardial patch reconstruction, and graft interposition having been described in the literature.[4,8] Repairing the aneurysm by plication or aneurysmorrhaphy is a quick and simple technique, but this can still place the patient at risk for a recurrent aneurysm later in life. Therefore, we used the tubular graft interposition technique in which the diseased arterial wall was completely excised because it offers superior long-term results.

The management of a pulmonary trunk aneurysm is a life-threatening issue, and there are no clear guidelines regarding the surgical indication for treatment. However, we believe that this pathology should be treated surgically with an acceptable risk of morbidity and mortality to prevent potentially catastrophic complications. In addition, we recommend elective surgical repair, especially when symptoms are present.

Declaration of conflicting interests

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