A Thrombotic Primary Venous Aneurysm of an Upper Extremity Causing Pulmonary Emboli after It Was Squeezed

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A 27-year-old Korean male complained of chest pain and dyspnea that began after he had squeezed a mass on the medial side of his right upper arm. Computed tomography angiography and venous Doppler ultrasonography revealed a right basilic vein aneurysm with an organized thrombosis causing pulmonary emboli. After 1 month of anticoagulation, the aneurysm was ligated and resected. During the operation, multiple venous aneurysms filled with organized thrombi were observed. This is the first reported case of a thrombotic primary venous aneurysm of an upper extremity causing pulmonary emboli after it was squeezed.

Venous aneurysms are rarely observed in the neck or upper extremities, and few symptoms other than aesthetic concerns have been reported.[1–7] Recently, 2 case studies described patients with venous aneurysms involving the neck and upper extremities, respectively, which provoked venous thrombosis and subsequent pulmonary embolisms.[8,9] Here, we describe a patient who experienced sudden onset of chest pain and dyspnea after he squeezed a mass in his right upper arm, which was confirmed to be venous aneurysm with thrombosis that caused a pulmonary embolism.

CASE REPORT

A 27-year-old Korean male visited the emergency department with sudden onset of chest pain and dyspnea. He also complained of a painless, compressible mass on the medial side of his right upper arm with adjacent swelling. He first noticed the mass during elementary school, and experienced intermittent dyspnea several times thereafter, but did not seek medical care. On the day of his hospital visit, he realized that his mass had become larger and hardened and squeezed it. The mass returned to its smaller size; however, he suddenly experienced chest pain and dyspnea, which required his transport to the emergency department. The medical and family histories were noncontributory, and he did not report any recent instance of extended sitting or lying supine. The patient also had not experienced any recent or past injury to his right upper arm, nor performed any repetitive arm motions or exercises in his daily life. Neither his right arm nor shoulder had ever been radiologically examined. On physical examination of his right upper arm, a soft, compressible, 8 × 5-cm, nontender mass was palpated; there was no overlying erythema or signs of phlebitis.

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Arterial blood gas analysis revealed that PaO_2 was decreased to less than 60 mm Hg, and d-dimer was elevated to 2.5 μg/mL.

Pulmonary computed tomography (CT) angiography revealed multifocal filling defects in the bilateral pulmonary arteries suggestive of pulmonary embolism (Fig. 1A–D). Brachial CT angiography of the right upper arm showed an aneurysmal dilatation of the basilic vein containing an acute thrombus on the proximal side (Fig. 1E), whereas femoral CT angiography revealed no evidence of deep vein thrombosis in the lower extremity (figure not shown). A color Doppler ultrasound examination confirmed that the basilic vein aneurysm was of a mixed nature (sluggish, turbulent venous blood flow from the proximal segment; Fig. 1F, G). Collectively, these findings suggest that a venous aneurysm involving partial thrombus in the upper arm was the source of the patient's acute pulmonary embolism.

We recommended a thrombophilia evaluation, but the patients refused because of the cost. Anticoagulation therapy with low-molecular-weight heparin (LMWH) was initiated on the day of admission. Specifically, 85.5 U/kg of nadroparin calcium was administered subcutaneously every 12 hr. We recommended surgically removing upper arm venous aneurysm and thromboses therein. However, the patient requested discharge as early as possible to return to his job. Therefore, we discharged him after his vital signs were confirmed to be stable, switching anticoagulation therapy from LMWH to rivaroxaban, an orally active direct factor Xa inhibitor on the seventh day of admission.

After 3 weeks of anticoagulation with rivaroxaban (15 mg twice daily), the patient requested that the aneurysm in his arm be surgically removed as soon as possible. On follow-up CT angiogram, we observed that the pulmonary embolism had resolved. He was readmitted and underwent surgery after being anticoagulated. During the operation, a longitudinal incision was made below the palpable mass and extended along the course of basilic vein in right upper arm. After gentle dissection to avoid a repeat pulmonary embolism, multiple saccular venous aneurysms ($60 \times 70 \times 60$ mm^3, $20 \times 25 \times 20$ mm^3, $25 \times 20 \times 15$ mm^3, and $15 \times 15 \times 10$ mm^3) were exposed. These appeared bead-like and were filled with fresh thrombi that could potentially embolize to the pulmonary arteries (Fig. 2A, B). The aneurysms were each ligated just above and below the aneurysms in the right basilic vein to prevent the recurrence of pulmonary embolism and excised. We observed that the thrombi were confined to within aneurysms. Finally, this was confirmed pathologically (Fig. 2C–E). Histologic examination revealed irregularly thickened and thinned vessel walls with decreased amounts of smooth muscle cells, increased fibrous tissue, and calcified thrombi, but no other abnormal findings. Notably, infiltration of inflammatory cells, degenerative changes, and arteriovenous fistulae indicating secondary causes of the aneurysm were all absent. The patient was discharged without complications or specific symptoms, and was given rivaroxaban for an additional 2 months.

DISCUSSION

A venous aneurysm is a marked, localized saccular or spindle-shaped dilation of a venous segment (i.e., at least 2.5–3 times the normal luminal diameter).[10]

The etiology can be classified as either primary or secondary.[11] The latter is caused by trauma, infections, venous valve insufficiency, or arteriovenous fistulas.[12] Primary aneurysms result from certain cardiopathies or venous pulmonary anomalies that decrease the blood flow to the right heart chambers.[13] In this case, there was no obvious primary or secondary cause, nor did the patient have a family history of venous aneurysm or thromboembolic event. The clinical presentation can vary widely.[14] Most patients complain of no more than local discomfort.[3] Venous aneurysms can cause pain and tenderness, but most are asymptomatic.[15] Many are initially misdiagnosed as soft tissue masses or inguinal or femoral hernias: some masses are painless, whereas others are painful and grow larger over time.[15,16] In this case, the venous aneurysm initially presented as a soft, compressible mass in the right upper arm that became larger and hardened over time, but was not accompanied by pain. On squeezing, it returned to its original size and texture. However, he experienced sudden-onset chest pain and dyspnea. As far as we know, there is no report of a pulmonary embolism caused by squeezing a thrombosed venous aneurysm.

Complications such as rupture, venous obstruction, mass effect, and thromboembolism are possible although most venous aneurysms are asymptomatic.[17–19] Venous aneurysms involving the lower extremities or intra-abdominal vessels are especially likely to require surgical intervention because of the high risk of thromboembolism.[1] Conversely, guidelines for the treatment of venous aneurysms of the upper extremities are not well established because of the paucity of cases.[9] Two of the 4 reported primary subclavian vein aneurysms led to surgical intervention (simple excision).[3–7] However, several aneurysms were saccular in nature, suggesting an increased likelihood of thrombosis compared with fusiform venous aneurysms. Recently, 2 case reports described venous aneurysms involving the neck and upper extremities that caused venous thrombosis and subsequent pulmonary embolism.[8,9] In 1 case, however, a pulmonary embolism was merely suspected, not diagnosed, because the