

Primary axillary venous aneurysm in a young patient presenting with cardiac arrest

Willythssa S. Pierre-Louis, MD,^a Raisa Tikhtman, BA,^b Alyssa Bonta, MD,^a and George Meier, MD,^a
Cincinnati, Ohio

ABSTRACT

Primary venous aneurysms are rare and usually asymptomatic. Venous aneurysms are manifested more frequently in the lower extremities than in the upper extremities. Primary venous aneurysms of the upper extremities are more often reported as aesthetically displeasing bulges or incidental findings. Here, we report the rare case of an axillary primary venous aneurysm in a pediatric patient who presented with syncope and massive pulmonary embolism and highlight the management. (*J Vasc Surg Cases and Innovative Techniques* 2019;5:375-8.)

Keywords: Venous aneurysm; Axillary aneurysm; Pediatric; Peripheral aneurysm

CASE REPORT

A 12-year-old white girl presented to an outside hospital with syncope and collapse during physical education class. Cardiopulmonary resuscitation was performed until the arrival of paramedics, who achieved return of spontaneous circulation en route to the emergency department. She had been complaining of left shoulder discomfort for 3 days before presentation. She denied a history of left arm swelling, numbness, or tingling as well as any prior trauma. She had no history of respiratory, cardiac, or hematologic disorders. There was no family history of aneurysm.

In the emergency department, she was hemodynamically stable although tachycardic. Physical examination revealed no evidence of a bulge or any skin changes. She denied tenderness in the left infraclavicular or axillary areas. Echocardiography was positive for pulmonary hypertension with right atrial and ventricular enlargement. Chest computed tomography angiography revealed a significant burden of pulmonary embolism originating in the branches of the main pulmonary arteries and extending distally (Fig 1). A 4.7- × 3.2- × 6-cm hyperattenuated collection in the left axilla adjacent to the axillary artery was visualized (Fig 2). This finding was read as an unknown mass or a hematoma. She underwent emergent placement of EKOS catheters (EKOS Corporation, Bothell, Wash) in bilateral pulmonary arteries and initiation of continuous tissue plasminogen activator and unfractionated

heparin. The EKOS catheters were removed on postoperative day 1 without complication, and she was transitioned from tissue plasminogen activator to a therapeutic heparin drip. The next day, venous duplex ultrasound ordered to find the source of the pulmonary embolism and to understand the computed tomography angiography findings revealed a thrombosed left axillary vein aneurysm (Fig 3). There were no large collaterals or involvement of the left brachial or subclavian veins. She was discharged 4 days later on therapeutic enoxaparin.

She was referred to our hospital for elective repair of the left axillary venous aneurysm 2 months after her admission. Repeated venous ultrasound examination demonstrated a recanalized aneurysm (Fig 3). We debated simply ligating the axillary vein aneurysm, which could result in limb swelling, vs aneurysmorrhaphy. If a bypass was needed, we would use the great saphenous vein and not a prosthetic for a conduit, given the patient's young age. We elected to perform an aneurysmorrhaphy by plication while she remained therapeutically anticoagulated with enoxaparin. An infraclavicular incision allowed access to the axillary vein along the deltopectoral groove. The dissection was guided by intraoperative ultrasound of the aneurysm and palpation of the artery. A normal segment of the vein was identified proximally. Dissection laterally revealed the bulging aneurysm. We minimized manipulation of the sac to limit the risk of embolization. It was dissected free circumferentially. The aneurysm sac was opened after proximal and distal control was obtained. The cavity was inspected; no thrombus was visualized, and a normal portion of the vein was demarcated from the weakened, pathologic aneurysmal wall (Fig 4). The inferior wall of the aneurysm was resected to normal tissue. With a running 6-0 Prolene suture, the normal layers were closed, leaving the superior flap of the aneurysm. This flap was then plicated and sutured onto the remaining inferior wall of the normal vein using another layer of continuous 6-0 Prolene. The patient tolerated the surgery well. Enoxaparin was continued, and she was discharged the next day. She has had normal findings on serial venous duplex ultrasound studies of the left upper extremity now 8 months after the repair. She denies any shoulder pain. The enoxaparin was discontinued after 3 months.

From the Division of Vascular Surgery, University of Cincinnati Medical Center^a, and the University of Cincinnati College of Medicine.^b

Author conflict of interest: none.

Correspondence: Willythssa S. Pierre-Louis, MD, Division of Vascular Surgery, University of Cincinnati Medical Center, 231 Albert Sabin Way, ML 0513, Cincinnati, OH 45267 (e-mail: pierrews@ucmail.uc.edu).

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Fig 1. Bilateral large pulmonary emboli originating in the main pulmonary artery branches and extending distally. A, Left axillary venous aneurysm; B, left axillary venous aneurysm.

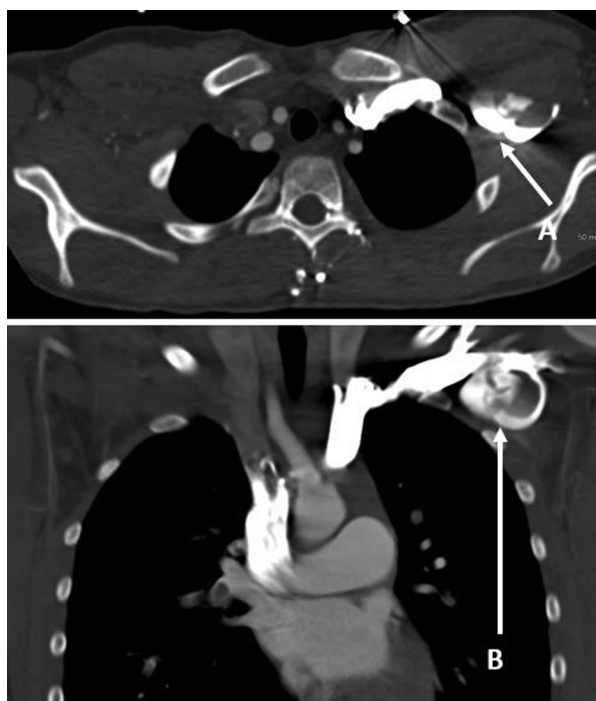


Fig 2. Left axillary venous aneurysm seen on axial (A) and coronal (B) computed tomography angiography of the chest.

Consent for publication was obtained from the patient's mother and father according to University of Cincinnati Medical Center protocols.

DISCUSSION

Venous aneurysms were first described by Sir William Osler in 1915.¹ The first case of pulmonary embolism from a thrombosed venous aneurysm was documented in 1976.² Venous aneurysms are defined as localized out-pouchings that communicate directly with the normal venous lumen with an anatomy distinct from varicose veins, arteriovenous malformations, and pseudoaneurysms.^{3,4} A consensus regarding the degree of dilation qualifying as aneurysmal does not exist. Definitions of

abnormal enlargements range from 1.5 to 3 times the baseline venous diameter.^{3,5-7}

Venous aneurysms are attributed to congenital or acquired deficiencies in connective tissue components of the venous wall. Another theory characterizes them as products of chronic venous insufficiency manifesting in "end-stage" varicose veins.⁸ Irwin et al⁹ observed increased expression of metalloproteinases, fragmented elastic lamellae, and loss of smooth muscle cells in eight venous aneurysm specimens compared with seven normal and varicose vein specimens. These findings reinforce that aneurysmal tissue is characterized by increased degradation and remodeling of the extracellular matrix.

Venous aneurysms of the head and neck or the upper extremities can be manifested as soft, painless, and compressible masses that enlarge with Valsalva maneuvers.⁵ In pediatric populations, the jugular vein is the most common site of aneurysm. Venous aneurysms are typically diagnosed in children secondary to a presumed congenital etiology, similar to our case.^{3,10} Diagnosis is made by venous duplex ultrasound, computed tomography, or venography. No individual method has been proven to be superior.³ The natural history of venous aneurysms of the upper extremity can be benign, with the aneurysmal tissue often being manifested as a nuisance.^{3,11}

The most feared complication of a venous aneurysm remains a pulmonary embolism, commonly associated with a popliteal vein aneurysm.^{3,5-7,12-14} In a case series of 31 patients, Calligaro et al¹⁴ noted that 71% of patients with deep venous aneurysm of the lower extremity experienced deep venous thrombosis or pulmonary embolism. Rupture of weakened venous aneurysmal tissue secondary to thrombotic obstruction represents another concerning sequela with the potential for fatal hemorrhage, although this complication has rarely been documented.^{12,14-17} A 2010 case report of a 40-year-old woman with a thrombosed external jugular vein aneurysm associated a primary venous aneurysm of the upper body with suspected pulmonary embolism.¹⁸ Furthermore, in 2013, Wallace et al¹⁹ reported the first case of a pulmonary embolism resulting from thrombosis of a proximal brachial-distal axillary venous aneurysm in their 22-year-old patient. Choi et al²⁰ described a patient who presented with symptomatic pulmonary embolism after he squeezed a painless mass felt in his upper arm, found to be a thrombosed basilic vein aneurysm.

Because of the risk of significant morbidity and mortality associated with aneurysms of the deep veins of the lower extremity, practice typically entails prophylactic surgery. Such a standard, however, has yet to be established for primary venous aneurysms of the upper extremity because of the scarcity with which symptomatic cases occur. Given the potentially fatal consequences, we recommend surgical management of upper extremity venous aneurysms over medical

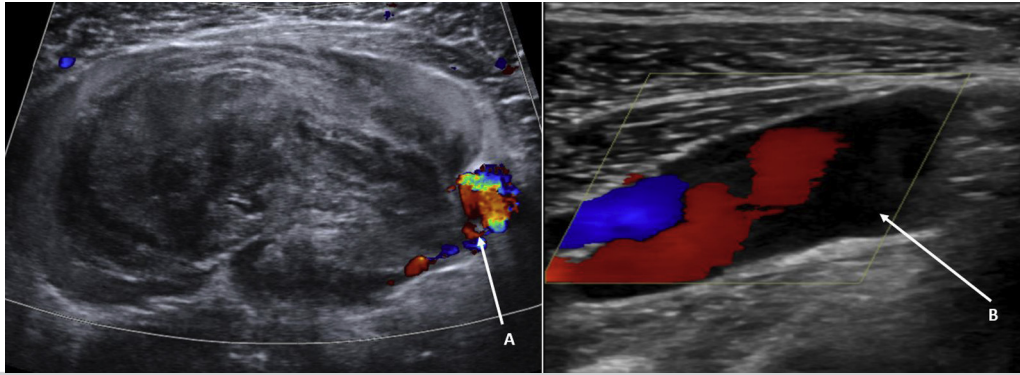


Fig 3. A, Thrombosed 6.1- × 3.0-cm left axillary venous aneurysm. B, Recanalized axillary venous aneurysm 2 months later.

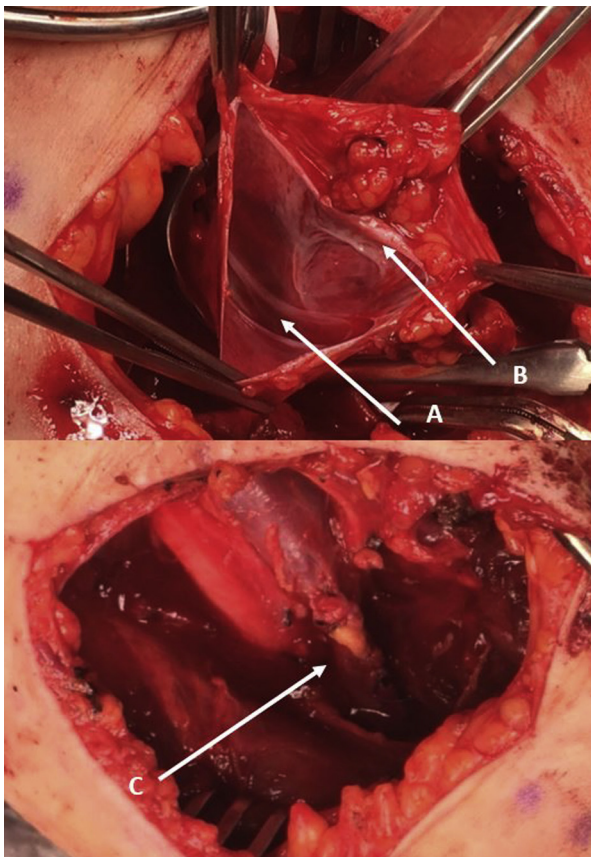


Fig 4. Opened aneurysm sac with normal venous tissue inferiorly (A) and aneurysmal wall (B). Repaired axillary vein after plication (C).

management (anticoagulation) or observation. Surgical options include ligation, interposition bypass, and aneurysmorrhaphy.

CONCLUSIONS

We report a rare presentation of a symptomatic primary venous aneurysm in a 12-year-old girl presenting with nearly fatal massive pulmonary embolism. Upper

extremity venous aneurysms are rare. Research is needed to help understand the etiology and natural history of this entity. Nevertheless, venous aneurysmorrhaphy remains safe and practical for the management of this potentially life-threatening problem.

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