Bilateral nonthrombotic subclavian vein obstruction causing upper extremity venous claudication

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Venous complications of thoracic outlet obstruction are frequently the result of acute axillosubclavian vein thrombosis, leading to symptoms consistent with venous claudication, including pain, swelling, and cyanotic discoloration. Nonthrombotic subclavian vein obstruction, however, is an uncommon cause of veno-occlusive symptoms. We report the case of a patient who, while running, developed pain consistent with venous claudication in her left arm and subsequently in her right arm. Clinical and hemodynamic evaluation revealed nonthrombotic subclavian vein obstruction, which was relieved by thoracic outlet decompression following first rib resection. (J Vasc Surg 2010;52:208-11.)

Thoracic outlet compression can cause neurogenic symptoms if the brachial plexus is compressed, arterial signs and symptoms if the subclavian artery is compromised, and veno-occlusive signs and symptoms with acute thrombosis if the subclavian vein is involved. However, it is unusual for patients to experience nonthrombotic veno-occlusive symptoms.

The superior and inferior limits of the thoracic outlet are the clavicle first rib and medially the subclavius tendon. The anterior scalene muscle is the lateral boundary for subclavian vein egress from the thoracic outlet. Most reports of venous complications of thoracic outlet (inlet) obstruction are the result of acute axillosubclavian vein thrombosis, leading to arm swelling, cyanotic discoloration, and distended subcutaneous collaterals. We report an interesting patient who had initial symptoms of pain in her left arm while running, consistent with venous claudication, followed by similar symptoms in her right arm. Subsequent evaluation demonstrated nonthrombotic subclavian vein obstruction, relieved by thoracic outlet decompression following first rib resection.

CASE REPORT

A 25-year-old athletic female was referred with symptoms of progressive edema and cyanosis of the left upper extremity during the preceding 2 to 3 months. During her waking hours, she developed tightness in her hands and especially her left arm, preventing her from wearing her wristwatch and rings. With exertion, her symptoms progressed to painful discomfort of her left arm, which was relieved with rest. Upon examination, she had mild edema of the left upper extremity with prominent superficial veins on the left anterior chest wall. Both radial pulses were palpable upon hyperabduction maneuvers of her arms. A duplex ultrasound was negative for deep vein thrombosis; however, there was diminished respiratory phasicity and a continuous flow signal in the left subclavian vein, consistent with venous obstruction.

She underwent bilateral upper extremity phlebography with dynamic maneuvers, demonstrating stenosis of the left subclavian vein as it crossed the first rib and prominent first rib collaterals, progressing to occlusion with hyperextension (Fig 1). The right subclavian vein occluded with hyperabduction but had extensive collaterals (Fig 2). The right arm was asymptomatic.

A left first rib resection was recommended, and the patient agreed. Through a transaxillary approach, the left thoracic outlet was exposed (Fig 3), and the left first rib was resected from the manubrium medially to the insertion of the middle scalene muscle laterally, posterior to the brachial plexus. On the second postoperative day, she underwent balloon venoplasty of the stenotic left subclavian vein (Fig 4), resulting in normal venous drainage. A stent was not used since there was no vein wall recoil with deflation of the angioplasty balloon. She was discharged to home on postoperative day three. Her left arm became asymptomatic.

During the next 2 years, she developed pressure, pulsatile discomfort, numbness, and overt pain in her right upper extremity, increasing in severity with physical activity. She complained of excruciating pain in her right arm when running, which caused her to stop. Initially, the pain was relieved by rest, but it progressed to the point that even at rest she experienced discomfort, discoloration, and cyanosis of her right arm and hand.

The patient's venous and arterial hemodynamics were studied with her arm in the neutral and hyperextended position at rest and after running on a treadmill until her right arm became painful. Air plethysmographic evaluation of the right arm at rest in the neutral position demonstrated a 1-second venous outflow fraction of 73%. In the abducted position, the outflow fraction dropped to 25%. Arterial inflow was 11.4 mL/min in the neutral position, which decreased to 9.2 mL/min in the hyperextended position. Following running on a treadmill, the 1-second outflow fraction dropped to 46% in the neutral position and 45% in the hyperextended position. The postexercise arterial inflow increased to 29 mL/min in the neutral position but dropped to 10.8 mL/min in the

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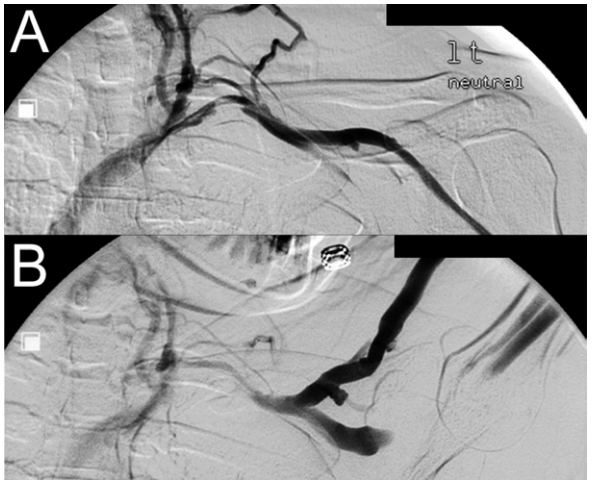

Fig 1. Phlebography of patient's symptomatic left upper extremity (**A**) reveals collateral development. (**B**) Upon abduction, occlusion of the subclavian vein is shown.

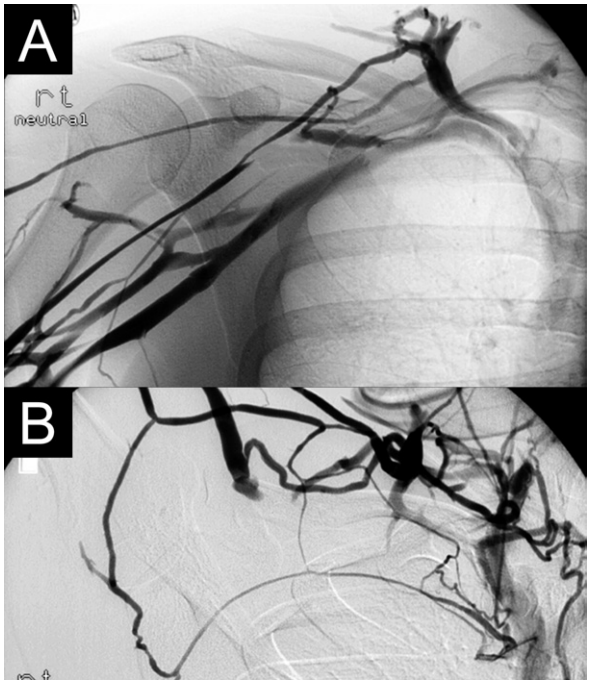

Fig 2. (**A**) Patient's right upper extremity was asymptomatic, but (**B**) abduction demonstrated occlusion of the subclavian vein with drainage from extensive collaterals.

hyperextended position. These physiologic studies confirmed significant compromise of the right subclavian vein leading to reductions in both venous outflow and arterial inflow, which were magnified following exercise.

She underwent repeat preoperative right upper extremity phlebography to rule out thrombosis and confirm the pathologic anatomy prior to decompression, which demonstrated fewer first rib collaterals than the phlebogram 2 years earlier (Fig 5). She then

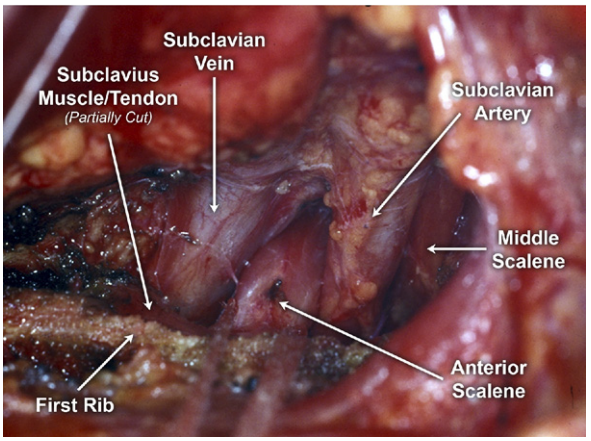

Fig 3. Operative photograph of the anatomy of the axillary space and thoracic outlet as seen through the transaxillary approach to first rib resection.

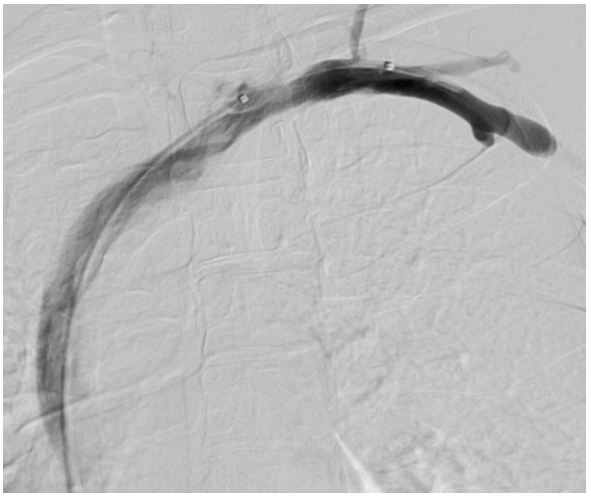

Fig 4. Venogram 1 day after left first rib resection following balloon dilation.

underwent transaxillary resection of her right first rib without complication.

She recovered well from her operation and was discharged home. One month later, she underwent bilateral upper extremity phlebography and had balloon angioplasty of her right subclavian vein, restoring normal venous drainage (Fig 5). Her left subclavian vein remained patent (Fig 6).

Six months after the procedure, she had an air plethysmographic evaluation of her right arm, which demonstrated a 1-second venous outflow fraction of 45.5% at rest, increasing to 52% in the abducted position. An arterial inflow study was performed, demonstrating an arterial inflow of 17.3 mL/min in the neutral position, which increased to 19.6 mL/min in the hyperextended position. These studies revealed significant improvement in arterial inflow and venous outflow following first rib resection. She remains asymptomatic at rest and with exercise.

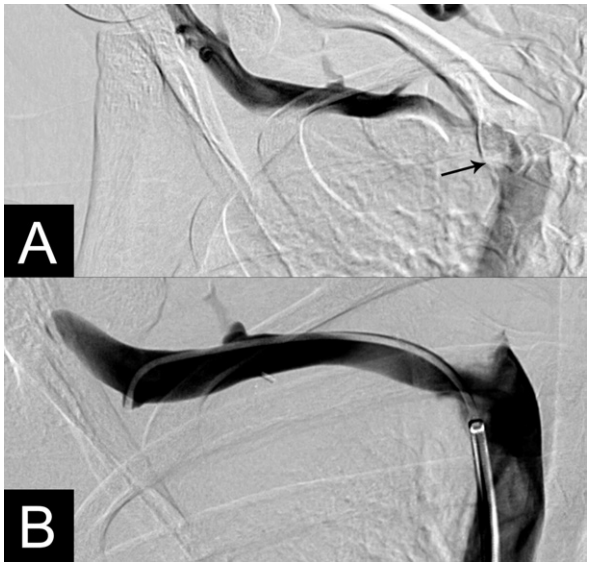

Fig 5. (**A**) Venogram showing preoperative right subclavian vein occlusion (arrow) in hyperabducted position and (**B**) normal anatomy with restored venous drainage when hyperabducted.

Fig 6. Venogram of left axillosubclavian veins 2 years following left first rib resection.

DISCUSSION

Our 25-year-old female patient had progressive symptoms of pain, edema, and cyanosis of her upper extremities, with increased upper extremity pain with running, typical of venous claudication due to nonthrombotic venous obstruction of her subclavian veins at the thoracic outlet. Physical findings were consistent with venous outflow obstruction confirmed by noninvasive physiological studies and phlebography. She underwent first rib resection to decompress both thoracic inlets and has done well since the procedures, with complete resolution of symptoms at rest and after exercise.

Table. Physiologic testing of the right upper extremity at rest and after exercise

	Resting		*Postexercise*	
Position	*1 sec MVO (%)*	*Arterial inflow (mL/min)*	*1 sec MVO (%)*	*Arterial inflow (mL/min)*
Preoperative				
Neutral	73.0	11.35	46.0	29.00
Hyperextended	25.0	9.20	45.0	10.80
Postoperative				
Neutral	45.5	17.30	45.5	38.60
Hyperextended	52.0	19.60	34.5	33.00

MVO, Maximal venous outflow.

Treatment of nonthrombotic subclavian vein obstruction was first reported in 1939 by McLaughlin et al.[1] Finding no venographic or operative evidence of thrombus, they noted narrowing of the subclavian vein where it crossed the first rib and performed a scalenotomy. The patient's symptoms resolved postoperatively. Subsequent cases of nonthrombotic subclavian vein obstruction have been reported and were recently summarized by Sanders et al.[2]

Our case is unique in regards to clinical presentation. Initially, our patient was asymptomatic at rest but developed symptoms only with exercise. Her pain symptoms fit the classic description of venous claudication, although affecting a nonexercising upper extremity. Her upper extremity motion during running likely compromised venous outflow, while, at the same time, the arterial inflow to her arms increased as a result of her increased cardiac output during running. Moreover, the venous obstruction prevented adequate venous drainage in the face of increased arterial inflow. Therefore, symptoms of bursting pain and swelling of her arm(s) occurred.

Physiologic testing of both venous and arterial hemodynamics in the neutral and hyperextended position, at rest and after treadmill exercise, demonstrated the compressive effect of the first rib on venous outflow and the restrictive effect of outflow obstruction on arterial inflow (Table). Paolini et al[3] reported that chronic venous obstruction in the lower extremities reduced arterial inflow at rest and following postocclusive hyperemia. We have demonstrated similar reduction in this patient's arterial inflow resulting from her proximal venous obstruction.

Air plethysmography (APG) is a volume plethysmograph that records the change in volume over time of the limb being evaluated. Maximal venous outflow evaluation records the rapidity of volume change in the limb following transient, partial venous occlusion. Arterial inflow is recorded as the volume change over time following transient arterial occlusion. With this patient's limb studied in the neutral and hyperextended positions, the hemodynamic effect of subclavian vein obstruction was objectively evaluated. These studies were repeated after the patient ran on a treadmill, both before and after her first rib resection, to

objectively correlate her hemodynamics with her symptoms preoperatively and her improvement postoperatively. While the APG added to our understanding of the pathophysiology of this patient's condition, the venographic findings were compelling and justified bilateral first rib resection.

This case summarizes the unusual presentation of upper extremity venous claudication with running, resulting from abnormal venous and arterial hemodynamics caused by nonthrombotic subclavian vein obstruction at the thoracic inlet, which normalized following first rib resection and subclavian vein venoplasty.

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