Effort Thrombosis in 2 Athletes Suspected of Musculoskeletal Injury

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Abstract

Objective: This case series describes the clinical presentation of effort thrombosis and the utility of sonography in its diagnosis.

Clinical Features: Two young male athletes presented to separate chiropractic clinics with suspected musculoskeletal shoulder injury. The first complained of dull shoulder pain after pitching in a baseball game and had no other signs or symptoms. The second presented after performing a weighted plank exercise and had prominent edema and discoloration of the affected extremity.

Intervention and Outcome: In the first patient, who had no physical signs to suggest thrombosis, a normal sonographic musculoskeletal shoulder exam prompted imaging in the abduction−external rotation position. This provided visualization of a thrombus in the axillosubclavian vein. Emergent referral followed, and treatment was initiated with thrombolysis and surgery, which resolved his condition. The second patient had physical signs consistent with effort thrombosis and was also referred to the emergency department, where sonography was performed and revealed thrombosis of the axillary and basilic veins. He responded to anticoagulants and thrombolysis and avoided surgery.

Conclusion: Effort thrombosis has a variable presentation that can mimic common musculoskeletal disorders and has a poorly defined diagnostic pathway. Acute shoulder pain in an athlete, especially with extremity edema after repetitive exertion, warrants urgent imaging such as sonography and emergent referral. Clinicians should understand the importance of a timely evaluation and diagnosis of effort thrombosis and that imaging may include sonography as a first-line imaging tool. (J Chiropr Med 2019;18:213-218)

Key Indexing Terms: *Effort-induced upper extremity deep vein thrombosis; Paget-Schroetter syndrome; Upper extremity deep vein thrombosis, primary; Venous thoracic outlet syndrome*

Introduction

Upper-extremity (UE) deep vein thrombosis (DVT) may be primary or secondary. Primary UE DVT can be further divided into idiopathic, venous thoracic outlet syndrome (vTOS), or effort thrombosis (ET).[1] Effort thrombosis, also known as Paget-Schroetter syndrome, is defined as axillary or subclavian vein thrombosis after strenuous upper limb activity.[2] It has a higher prevalence in young, otherwise healthy active individuals[2] and has a male-to-female prevalence of 2 to1.[3] About 60% to 80% of cases occur after vigorous overhead activity,[4] whereas other causes include heavy lifting[5] and manual labor.[6] Effort thrombosis is rare, with an incidence of 1 to 2 per 100 000 people/y[3] compared to 91 per 100 000/y for lower-extremity DVT.[7] Secondary DVT of the UE results from a known cause such as an implanted central venous catheter[8] and is relatively more common than primary, occurring at a rate of 14 per 100 000/y.[7]

The clinical examination is rarely specific enough to rule in UE DVT, requiring that further testing be performed when it is suspected.[9] Sonography has emerged as a recommended first test for the detection of UE DVT,[10] and the utility of initial d-dimer testing for suspected UE DVT is under investigation.[9] When these are inconclusive, further imaging may be sought when clinical suspicion is high, but the diagnostic pathway is poorly defined in cases lacking obvious clinical signs.[11-13] Timely diagnosis and treatment of ET avoids potential complications, such as pulmonary thromboembolism and post-thrombotic syndrome of the arm.[11,14,15] We present 2 cases that highlight the variable clinical presentation of ET and the use of ultrasound (US) as a first-line imaging test. The patients discussed in the following case reports gave written consent for use of deidentified images and clinical data.

Case Reports

Case 1

A 14-year-old right-handed male patient presented with his parents to a chiropractor for evaluation of acute right

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shoulder pain after pitching in a baseball game. The patient was referred for US with suspicion of subscapularis tendon pathology, but no physical exam findings were received from the referring clinician. He underwent US within a week in the radiology department of a chiropractic teaching clinic. He was imaged (GE Logiq E9, GE Healthcare, Wauwatosa, Wisconsin) using 2 high-frequency linear transducers (ML6-15 operating at 12 MHz and L8-18i operating at 15 MHz).

Sonographic examination of the right shoulder using a standardized musculoskeletal (MSK) protocol[16,17] revealed no abnormal findings. Specifically, there was a normal appearance of the subscapularis tendon, which was the anatomy of interest and directed the imaging examination. Because there were no abnormal findings on the initial examination, additional images were obtained in the area of greatest pain intensity as indicated by the patient. This was in the general area of the insertion of the subscapularis tendon, but near the axilla. Therefore, the patient's arm was repositioned, and an add-on US protocol was initiated.[18]

Examination in the abducted, externally rotated arm position (Fig 1) identified thrombosis of the subclavian vein extending into the axillary vein that correlated with the area of chief complaint (Fig 2). That arm position was sought specifically to inspect the insertion of the latissimus dorsi tendon[19] based on the location of pain, and the thrombus was an incidental finding that was unexpected. Visual inspection of the upper extremity failed to yield evidence of superficial collateral veins, cyanosis, or edema.

Fig 1. *Clinical example of the abducted, externally rotated position of the shoulder during sonographic examination. This position was used in case 1 to identify a thrombus that was not visualized in the standard examination.*

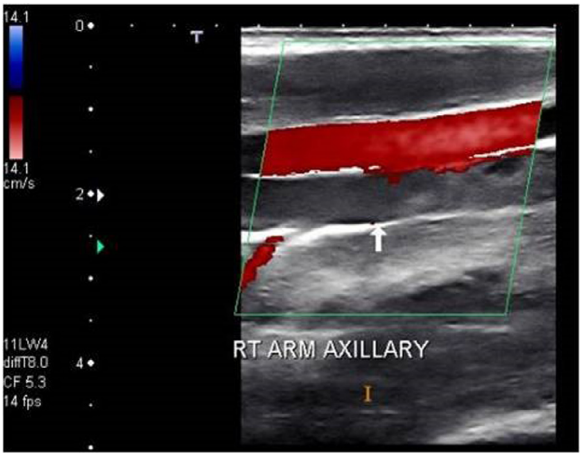

Fig 2. *Sonographic imaging (long axis) using color Doppler demonstrating flow within the axillary artery, but not within the axillary vein (arrow), of our case 1 patient. The echogenic content within the venous lumen is thrombus.*

Subsequent to the US examination, emergent referral was initiated with a preliminary diagnosis of DVT in the axillosubclavian veins. The diagnosis of UE DVT was confirmed by duplex US exam using compression technique at a nearby emergency department. The vascular protocol used also targeted the axillosubclavian area, where the thrombus was confirmed. Initial treatment consisted of pharmacologic or catheter-directed thrombolytic therapy. Subsequent US imaging revealed persistent thrombosis, after which surgical thrombectomy and first rib removal were performed without incident.

After the intervention, the patient reported no complications. At 6 months' follow-up, he had fully recovered from surgery, was pain free, and returned to full participation in sports, including pitching.

Case 2

A 28-year-old right-handed male fitness instructor presented to a chiropractor with a 2-day history of frequent moderate pain and paresthesia of the entire left upper extremity and hand. Symptoms began while doing a vigorous exercise routine in which he was holding a prone abdominal plank position for over-1-minute repetitions with a 45-pound plate fixed to his back. He noted maximal pain around the left clavicular and axillary regions, and increased symptoms when raising the left arm overhead. The patient was a nonsmoker and denied use of anabolic steroids. He reported a family history of factor V Leiden mutation in his mother. Also, both of his grandfathers suffered from strokes, the first at age 35 (paternal) and 67 (maternal) and the second, fatal stroke, both at age 77. He denied any history of trauma to the left shoulder or arm, but thought he strained his shoulder muscles during the plank workout.

Fig 3. *Diffuse swelling and cyanotic discoloration of the left upper extremity, suggestive of proximal thrombosis seen in case 2. In addition, the cephalic vein of the left mid-arm appears engorged.*

Examination revealed diffuse reddish-blue discoloration, warmth, and non-pitting edema of the left upper extremity from the proximal arm to the hand (Fig 3). Collateral subcutaneous veins were visible around the left anterior shoulder, a feature of UE DVT termed Urschel's sign[20] (Fig 4). Palpation of the axilla yielded a hard and tender linear cord, a sign of axillary vein thrombosis.[6] Brachial and radial pulses and capillary refill were normal. No other testing maneuvers for thoracic outlet syndrome were performed.

The patient was suspect for a thrombosis and was referred to the emergency department. The hospital staff was informed of the suspicion of ET. A duplex venous US exam confirmed thrombosis of the left axillary vein extending into a brachial vein, accompanied by a superficial thrombus of the basilic vein.

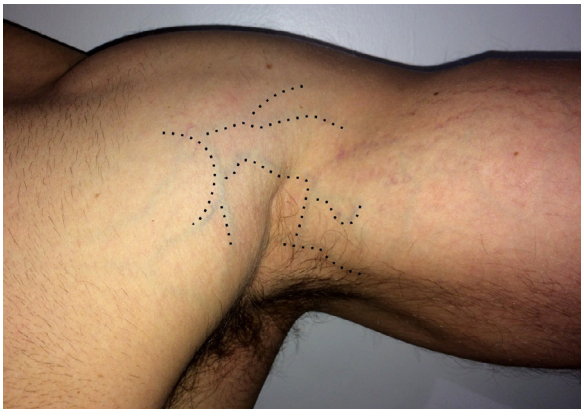

Fig 4. *Dilated collateral subcutaneous veins (dotted lines) at the left anterior shoulder (case 2). This sign of effort thrombosis is called Urschel's sign. Arm cyanosis is also visible.*

The patient was given anticoagulants (rivaroxaban), and the following week had a venogram with subsequent pharmacologic/catheter-directed thrombolytic therapy. The vascular surgeon prescribed a 6-month course of anticoagulation. Upper-extremity edema and discoloration diminished significantly after thrombolysis with the remainder gradually subsiding over the following 2 months. The patient was informed of the option for surgical decompression but declined. The patient indicated satisfaction with the quality of his pharmacologic outcome and was able to modify workouts, avoiding overhead exercise and planks.

Discussion

Deep vein thrombosis of the UE is classified as primary or secondary. Secondary UE DVT is far more common (80% of cases) and is caused mainly by indwelling intravenous catheters, but also by pacemakers, malignancy, or thrombophilia.[12] Primary UE DVT is less common and may be classified as idiopathic, vTOS, or ET.[1] Effort thrombosis, also known as Paget-Schroetter syndrome, occurs after strenuous UE activity and may be thought of as a consequence of vTOS.[2,13,21] Those with vTOS have risk factors for compression of vascular structures in the costoclavicular space including cervical ribs, fibrous bands, scalene or subclavius muscle hypertrophy, and anomalous insertion of the costoclavicular ligament.[2,3] The axillary and subclavian vessels may additionally undergo strain in the end-range position of throwing a baseball overhead (hyperabduction, extension, and external rotation of the humerus).[2,21-23]

Although ET has been most extensively documented in baseball players,[21,23] it may also occur after vigorous exercise in weight lifting[3,6] and bench pressing.[4] To our knowledge, ET has not been reported after plank exercises. In a plank exercise, the shoulder is adducted and depressed, which contrasts with the abducted and elevated position in throwing a baseball.

Throwing, as in case 1, is thought to repeatedly strain the subclavian and axillary veins and cause endothelial microtrauma and inflammation, leading to thrombosis.[1,2,15,21,24] In comparison, weight lifting causes hypertrophy of the subscapularis, pectoralis minor, scalene, and subclavius muscles, and may compress the subclavian vein in the thoracic outlet and cause thrombosis.[6] In case 2, the onset of symptoms after prolonged, repeated shoulder exertion in an adducted position during plank exercises is suggestive of venous compression by muscle hypertrophy. Our cases exemplify 2 different mechanisms of ET, case 1 showing strain of the axillosubclavian veins, with case 2 primarily displaying compression without strain.

Upper-extremity edema and pain are the most common symptoms of ET,[2] but edema is not present in all cases.[11,21] Other signs and symptoms include a subjective sense of

arm heaviness[2,6] and palpation of a hard linear structure in the axilla.[6] Studies of patients with non-effort thrombosis UE DVT have identified a prevalence of UE swelling or pitting edema ranging from 70% to 87%.[9,25] One retrospective study reported a 93% prevalence of edema in a young athletic population with ET.[26] Absence of edema and other symptoms may be explained by compensatory venous collateral formation around the axillosubclavian vein.[11] Cases of ET similar to case 1 presenting to a chiropractic clinic without UE swelling have been published.[27,28] It is possible that cases of ET without swelling are more likely to masquerade as MSK pathology, causing patients to not seek emergency care.

Effort thrombosis has a broad differential that includes muscle injury, quadrilateral space syndrome, lymphangitis, lymphedema, hematoma, superficial vein thrombosis, skin infection, neurogenic or arterial TOS,[6,13,26,27] and rhabdomyolysis.[29] Overhead-throwing athletes without overt vascular signs and symptoms may be suspect for rotator cuff pathology or subacromial impingement, whereas those with pain and extremity swelling may be suspect for a vascular, inflammatory, or lymphatic disorder.[27]

Clinical decision rules help determine the probability of UE DVT. The Constans score has a sensitivity of 38% to 86% and specificity of 64% to 93%[9,25] and considers UE DVT likely with a score of $\geq$2 points for presence of a central venous catheter or pacemaker (+1), unilateral edema (+1), localized pain (+1), and another diagnosis being at least as plausible (−1).[25] An extended Constans score considers UE DVT likely with $\geq$3 points and additionally includes superficial vein dilation (+1), arm swelling > 1.5 cm more than the uninvolved side (+1), estrogen use or skin color difference (+1), and intravenous injection or peripheral venous catheter in the past 5 days (−1).[9] The extended Constans score has a sensitivity of 90% and specificity of 64%.[9] The patient in case 1 did not present with UE edema, superficial vein dilation, or skin color difference, and in retrospect would have had Constans and extended Constans scores of 0 (+1 for pain, −1 for MSK diagnosis being plausible). Because a low clinical probability does not rule out UE DVT,[25] clinicians must consider risk factors for ET such as recent vigorous arm exertion when deciding to refer for urgent imaging such as US.[2,3,21]

Duplex US has emerged as the first-line imaging tool for clinically suspected DVT.[8,13] Unlike venography, US is noninvasive, lacks ionizing radiation, and avoids potential complications associated with contrast.[13,30] For diagnosis of UE DVT, compression US demonstrated a sensitivity of 97% and specificity of 96%, and combined compression/Doppler US demonstrated a sensitivity of 91% and specificity of 93%.[30] One disadvantage of US imaging is its field of view. It is limited proximally, deep to the clavicle, which is a common location for thrombosis.[5,31] In those cases, venography is the next step in suspected UE DVT[3,13] and can be both diagnostic and therapeutic,[3,5] although other types of imaging are available as well.[32] Another potential disadvantage is that US is user dependent.[3,10] An inexperienced and less competent examiner could have missed the diagnosis in case 1. Although US is the first-line imaging choice for UE DVT, a negative US exam should be interpreted with caution owing to these limitations, especially when clinical suspicion is high.[11,33]

Complications of ET include pulmonary thromboembolism and post-thrombotic syndrome.[7] Pulmonary embolism is reported in 3% to 36% of UE DVT cases,[34] whereas post-thrombotic syndrome develops in 7% to 46%.[35] Symptoms of post-thrombotic syndrome include limb pain and swelling, heaviness, cramping, and edema with significant reduction in quality of life, especially in the dominant arm.[14,15] Pulmonary embolism can be asymptomatic or present with dizziness and shortness of breath, and has a potentially fatal outcome.[11,34] Recognition of the clinical signs and symptoms of ET and early imaging with US may help avoid these complications.

Catheter-directed thrombolysis combined with anticoagulation is the standard-of-care treatment for UE DVT, but has been questioned.[1,15] One recent systematic review found insufficient evidence to support the benefits or harms of thrombolysis, either alone or combined with other interventions.[36] Studies on proximal lower-extremity DVT have come to a similar conclusion, as a randomized clinical trial found that pharmacomechanical catheter-directed thrombolysis did not prevent occurrence of post-thrombotic syndrome at 6 to 24 months' follow-up.[37] Newer treatment options may be forthcoming, as US elastography may determine the age of a thrombus, influencing both the treatment approach and prognosis.[38]

It is generally accepted that anticoagulation therapy alone is insufficient and should be combined with surgical thrombectomy and thoracic outlet decompression for the best outcome.[11,14,15] The use of surgical decompression is based more on clinical experience and consensus rather than randomized controlled trials.[1,15] Those that choose a nonsurgical approach, such as in case 2, may be forced to restrict provocative activities and also should have ongoing anticoagulation therapy due to the risk for recurrent thrombosis.[5]

Limitations

There were limitations to this case report. Generalization of the diagnostic findings and treatments represented in this case series will not necessarily apply to a larger population. Also, further clinical findings in case 1 were not available to the authors.

Conclusion

We presented 2 cases highlighting the variable clinical presentation of ET, which should be considered in the

differential diagnosis of a young, overhead athlete with shoulder pain with or without swelling.[34] The variability of our cases shows that overhead and non-overhead athletes can be affected by ET, and clinical signs can be pronounced or not present at all. Patients with suspected ET should be emergently evaluated, and imaging protocols may include duplex vascular US.

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Contributorship Information

Concept development (provided idea for the research): R.M., R.J.T., N.W.K.
Design (planned the methods to generate the results): R.M., R.J.T., N.W.K.
Supervision (provided oversight, responsible for organization and implementation, writing of the manuscript): R.M., R.J.T., N.W.K.
Data collection/processing (responsible for experiments, patient management, organization, or reporting data): R.M., R.J.T., N.W.K.
Analysis/interpretation (responsible for statistical analysis, evaluation, and presentation of the results): R.M., R.J.T., N.W.K.
Literature search (performed the literature search): R.M., R.J.T., N.W.K.
Writing (responsible for writing a substantive part of the manuscript): R.M., R.J.T., N.W.K.
Critical review (revised manuscript for intellectual content, this does not relate to spelling and grammar checking): R.M., R.J.T., N.W.K.

Practical Applications

- An athlete presenting with arm cyanosis, edema, and prominent superficial veins after vigorous upper extremity overuse should be suspect for effort thrombosis.
- Athletes without visible signs of vascular pathology still may have effort thrombosis.
- Those with potential effort thrombosis should be referred for imaging such as US.
- A high index of suspicion and urgent decision-making is required to prevent the negative sequelae of this condition

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