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CASE REPORT

Venous Thoracic Outlet Syndrome Caused by Double Compression of the Axillosubclavian Vein: A Case Report

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Introduction: In venous thoracic outlet syndrome (VTOS), pathology around the axillosubclavian vein causes venous compression with the subsequent development of upper extremity symptoms. This case report describes the analysis of all possible compression sites and subsequent treatment of VTOS patients with multiple compression points.
Report: A 22 year old male presented with severe pain and swelling in his right arm, which persisted after a conservatively managed primary upper extremity deep vein thrombosis. Compression of the axillosubclavian vein was seen both at the level of the pectoralis minor and the costoclavicular spaces. Both compression points were successfully treated by combining thoracic outlet decompression surgery with pectoralis minor tenotomy.
Discussion: This report underlines the importance of considering the possibility of multiple compression sites in patients with VTOS. Incomplete surgical release of all compression points leaves patients prone to re-thrombosis and/or persistent post-thrombotic syndrome. Timely recognition of all abnormalities on venography may allow for adjustment of surgical treatment accordingly.

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BACKGROUND

In venous thoracic outlet syndrome (VTOS), pathology around the axillosubclavian vein causes venous compression with subsequent development of upper extremity symptoms. In general, VTOS clinically manifests as either intermittent positional obstruction, or more commonly, primary upper extremity deep vein thrombosis (UEDVT). The latter is also called "effort thrombosis" or Paget-Schroetter syndrome (PSS) and is a rare condition with an occurrence rate of approximately 1–2/100 000 people per year.[1]

Classically, primary UEDVT is either idiopathic or caused by compression of the axillosubclavian vein localised at the costoclavicular junction. This report presents a patient who appeared to have a double compression of the axillosubclavian vein; both in the costoclavicular space and in the pectoralis minor space. The possibility of multiple compression sites is well known in neurogenic thoracic outlet syndrome (NTOS).[2] In NTOS patients, analysis of the anatomy and subsequent correction of multiple compression sites has become the standard of care in recent years. However, multiple compression sites in VTOS patients are rare and consequently analysis of all potential compression points is not always conducted. Nevertheless, prolonged exposure to compression by the causative anatomy results in a cycle of inflammation and fibrosis of both the vein wall itself and adjacent structures, which ultimately can result in axillosubclavian vein thrombosis.[1] Therefore, in depth analysis of the anatomy in VTOS patients to identify and treat multiple compression sites can help to prevent long term sequelae. This case report describes the analysis of all possible compression sites and subsequent treatment of VTOS patients with multiple compression points.

CASE PRESENTATION

A 22 year old male presented to his general practitioner (GP) with severe pain and swelling in his dominant right arm. These complaints first appeared after carrying the coffin at his grandmother's funeral with both his arms abducted to around 90°. His medical history was unremarkable, apart from a fractured clavicle during birth (side unknown). No venous thrombo-embolic disease risk factors

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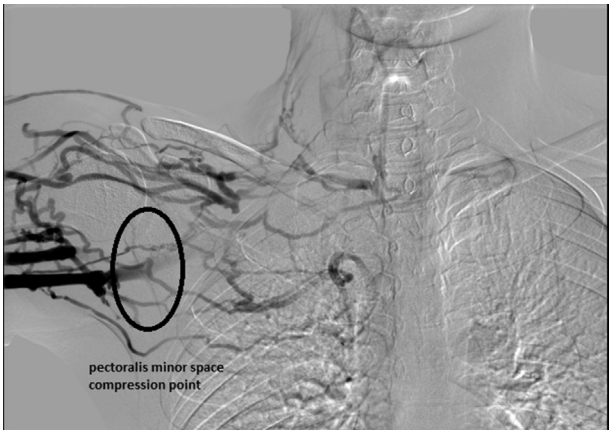

Figure 1. Arm at 90° abduction at presentation. A compressed axillary vein in abduction, situated at the lateral border of the pectoralis minor muscle with prominent collateral veins is seen in the right arm and thoracic outlet.

were present. Physical examination performed by the GP revealed a swollen, purple-red discoloured right arm with several distended veins. Computed tomography venography (CTV) at a local hospital revealed an UEDVT with total occlusion of the right subclavian vein. Initial therapy consisted of low molecular weight heparin (Nadroparin, Fraxodi, GlaxoSmithKline, Brentford, UK) and a vitamin K antagonist (acenocoumarol). The patient was advised to continue with acenocoumarol alone as anticoagulant therapy after hospital discharge. Nine months later, he presented again to the GP with persisting symptoms, severely limiting his daily activities. He was referred to the study centre because of the regional expertise with management of thoracic outlet syndrome.

Examination and imaging

The patient presented in the vascular surgery outpatient clinic with post-thrombotic syndrome (PTS) of his right arm and hand despite 10 months of oral anticoagulant therapy. Exercise provoked progressive oedema and pain, both alleviated with rest. These complaints necessitated wearing a compression sleeve, which partly reduced the symptoms. His symptoms severely limited his daily activities, especially after returning to work as a high school physical trainer.

Physical examination confirmed a full range of movements of the shoulder joint. However, prominent distended veins were visible in both the right shoulder region and proximal right arm. The elevated arm stress test (EAST) was positive after five seconds with complaints of heaviness and paraesthesia in the right arm. Screening for thrombophilia was conducted in the referring hospital and showed no abnormal findings. The Xray of the superior thoracic aperture showed no abnormalities. Duplex ultrasonography at rest revealed a patent axillosubclavian vein. However, in 100° abduction, a possible compression of the vein was seen both at the level of the pectoralis minor and costoclavicular spaces. As part of the standard work up in VTOS, venography was performed at rest, abduction, adduction,

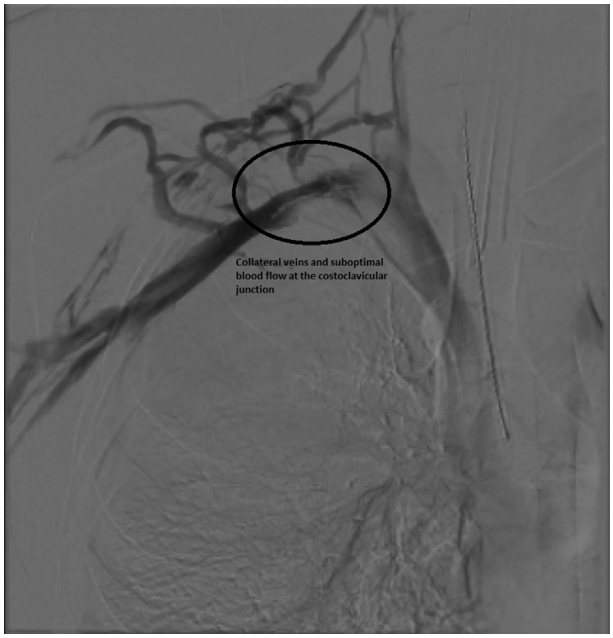

Figure 2. Situation directly after first rib resection in the operating room. The patency is restored at the level of the axillosubclavian vein. However, persistent collateral veins and suboptimal blood flow were seen in abduction at the level of the costoclavicular junction.

and military attitude. Venography showed a compressed axillary vein in abduction, situated at the lateral border of the pectoralis minor muscle with prominent collateral veins (Fig. 1). Also, more collateral veins were seen in the costoclavicular space confirming the suspicion of a second compression point.

Treatment. To adequately decompress both obstructions, transaxillary pectoralis minor tenotomy was performed at the level of the coracoid process as well as a thoracic outlet decompression (TOD) consisting of a complete first rib resection, partial anterior and medial scalenectomy, removal of the medial part of the subclavius muscle and a thorough, circumferential venolysis. In the same session, venography was performed in the hybrid operating room to assess vein patency (Fig. 2). The venogram revealed a positive effect of the surgery with patency restored at the level of the axillosubclavian vein. However, persistent collateral veins and suboptimal blood flow was still seen in abduction at the level of the costoclavicular junction. Passage of the residual lesion with a guide wire (0.035″ Glidewire, Terumo Medical Corp., Somerset, NJ, USA) was performed followed by a percutaneous transluminal angioplasty (PTA) with a 12×40 mm balloon (Powerflex, Cordis, Miami, FL, USA). After PTA, the axillosubclavian vein was then fully patent without collateral veins (Fig. 3).

Follow up. The patient was discharged on the first postoperative day with a prescription for six weeks of rivaroxaban 20mg once a day (Xarelto, Bayer, Leverkusen, Germany). Follow up venography at three and 12 months showed a patent axillosubclavian vein with no collaterals

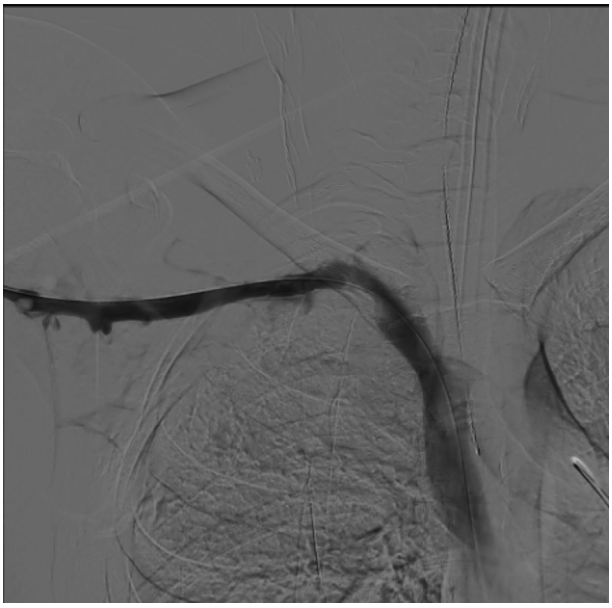

Figure 3. Situation after first rib resection and PTA in 90° abduction. The axillosubclavian vein was fully patent without collateral veins.

(Fig. 4). Moreover, the patient has had no recurrent UEDVT with over three years of follow up and experiences only some minor complaints in his arm during strenuous exercise.

DISCUSSION

Treatment modalities for PSS can be divided into two strategies: conservative treatment with anticoagulation and invasive treatment with thrombolysis in combination with TOD and venous reconstruction or PTA.[1,3] Although level 1 evidence is lacking, some experts favour invasive management including preliminary anticoagulation therapy, catheter directed thrombolysis, TOD surgery, and venous reconstruction or PTA.[1] This approach produces long term symptomatic relief in 84—95% of the patients.[1,4] In contrast, initial conservative management with (oral) anticoagulants is associated with long term morbidity, the risk of re-thrombosis, and pulmonary embolism.[5] Long term (18—60 months) disability rates in treatment pathways including TOD are significantly lower compared with conservative treatment pathways.[6,7] Initial conservative treatment in the present patient led to sequelae persisting for more than one year. Nevertheless, TOD surgery and PTA resulted in excellent short and mid term outcomes.

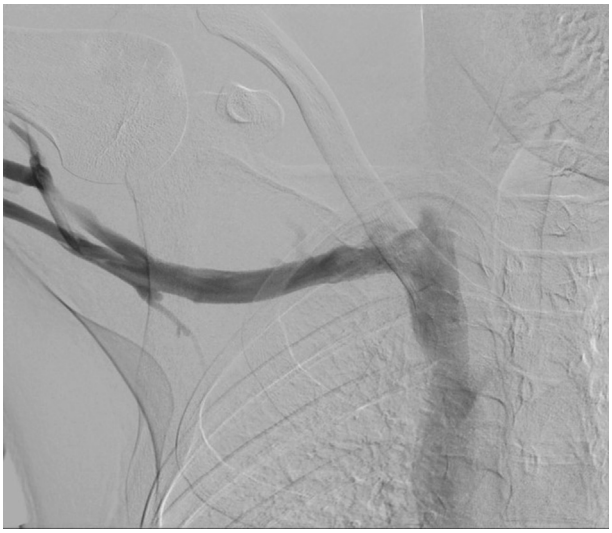

Figure 4. Follow up venogram after 12 months in 100° abduction, which shows a patent axillosubclavian vein with no collaterals.

In most patients with PSS, venous obstruction occurs solely in the costoclavicular space.[8] One of the anatomical abnormalities found in PSS is a more laterally inserted costoclavicular ligament compared with healthy individuals. This combined with a hypertrophied anterior scalene muscle leads to significant narrowing of the vein resulting in a cycle of inflammation and fibrosis of the vein wall itself and adjacent structures.[1] The resultant decreased blood flow through the axillosubclavian vein can consequently trigger the development of thrombosis as seen in patients with PSS.[9]

In the present case, the axillary vein was also compressed at the pectoralis minor space. Compression of the neurovascular bundle in the pectoralis minor space is rare in venous TOS, although more often seen in patients with neurogenic thoracic outlet syndrome (NTOS).[2] Obstruction of the axillary vein by the pectoralis minor muscle is even more unusual, with only 10 cases reported to date. In this case, compression in the pectoralis minor space was identified by seeing a more laterally compressed axillosubclavian vein compared with patients with costoclavicular space compression alone. Where compression of the axillosubclavian vein is a rare entity in the costoclavicular space and even more so at the level of the pectoralis minor space, a combination of obstruction at both sites in the same patient ('venous double crush syndrome') has not been described previously, to the present authors' knowledge.

It is important to define the level of obstruction to perform adequate TOD. Inadequate TOD surgery can lead to continued venous compression, making the patient prone to re-thrombosis and/or persistent post-thrombotic syndrome. Patients with an isolated obstruction in the pectoralis minor space can be treated with a relatively low risk pectoralis minor tenotomy (PMT) in day care surgery, as performed for NTOS.[10] In PMT 3—4 cm of pectoralis minor is removed routinely, so partial pectoralis minor resection better describes the procedure.

As demonstrated in this patient, venography allows for dynamic assessment of the vein with provocation of possible compression in at risk areas. Venography is used routinely with the arm at rest, in adduction, abduction and in a military attitude position. Using this imaging modality, it was possible to identify the presence of both compression sites and subsequently guide treatment. A transaxillary approach allows for both PMT and TOD with venolysis through the same incision. However, an infra- or paraclavicular approach with subsequent incision for a PMT would also suffice.

CONCLUSION

This case report describes the successful treatment of a double crush venous TOS in a patient with an upper extremity post-thrombotic syndrome caused by axillosubclavian vein compression at both the costoclavicular and pectoralis minor spaces.

It underlines the importance for clinicians to consider the possibility of multiple compression sites in patients with VTOS. Timely recognition of all abnormalities on venography may allow for adjustment of surgical treatment accordingly.

DECLARATION OF COMPETING INTEREST

None.

FUNDING

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REFERENCES

1 Illig KA, Doyle AJ. A comprehensive review of Paget-Schroetter syndrome. *J Vasc Surg* 2010;**51**:1538—47.
2 Upton AR, McComas AJ. The double crush in nerve entrapment syndromes. *Lancet* 1973;**2**:359—62.
3 Van den Houten MML, Van Grinsven R, Pouwels S, Yo LSF, Van Sambeek MRHM, Teijink JAW. Treatment of upper-extremity outflow thrombosis. *Phlebology* 2016;**31**:28—33.
4 Doyle A, Wolford HY, Davies MG, Adams JT, Singh MJ, Saad WE, et al. Management of effort thrombosis of the subclavian vein: today's treatment. *Ann Vasc Surg* 2007;**21**:723—9.
5 Vazquez FJ, Paulin P, Poodts D, Gandara E. Preferred management of primary deep arm vein thrombosis. *Eur J Vasc Endovasc Surg* 2017;**53**:744—51.
6 Urschel Jr HA, Razzuk MA. Paget-Schroetter syndrome: what is the best management? *Ann Thorac Surg* 2000;**69**:1663—8.
7 Elman EE, Kahn SR. The post-thrombotic syndrome after upper extremity deep venous thrombosis in adults: a systematic review. *Thromb Res* 2006;**117**:609—14.
8 Urschel Jr HC, Pool JM, Patel AN. Anatomy and pathophysiology of VTOS. In: Illig KA, Thompson RW, Freischlag JA, Donahue DM, Jordan SE, Edgelow PI, editors. *Thoracic outlet syndrome*. London: Springer-Verlag; 2013. p. 339—43.
9 Kucher N. Clinical practice. Deep-vein thrombosis of the upper extremities. *N Engl J Med* 2011;**364**:861—9.
10 Sanders RJ, Rao NM. Pectoralis minor obstruction of the axillary vein: report of six patients. *J Vasc Surg* 2007;**45**:1206—11.