

CASE REPORT

Infrequent origin of a peripheral embolism: Arterial thoracic outlet syndrome in a young woman

Periferik embolinin az rastlanan nedeni: Genç kadın hastada arteriyel torasik çıkış sendromu

✉ Berhan Keskin, M.D., ✉ İsmail Balaban, M.D., ✉ Seda Tanyeri, M.D.,
✉ Ahmet Karaduman, M.D., ✉ Ali Karagöz, M.D.

Department of Cardiology, Kartal Koşuyolu Heart Training and Research Hospital, İstanbul, Turkey

Summary– Arterial thoracic outlet syndrome (TOS) is caused by the compression of the subclavian or axillary artery between the ribs and the scalene muscles. Patients may present with various clinical conditions. Herein, we report a case of arterial TOS who presented with peripheral embolism. Arterial TOS is an uncommon cause of peripheral embolism. The diagnostic approach and treatment methods used are discussed. Clinicians should keep this diagnosis in mind and manage these cases according to the clinical appearance.

Arterial thoracic outlet syndrome (TOS) is caused by compression of the subclavian and/or axillary arteries between the ribs and the scalene muscles. A cervical rib is often involved. Arterial TOS is associated with subclavian aneurysms and blood clots.^[1] Patients may remain asymptomatic until ischemic symptoms are exacerbated by repetitive trauma or acute thrombosis.^[2] The medical history, physical examination, and imaging are crucial in the diagnosis. This is the case of a young female diagnosed with arterial TOS managed using a combined pharmacological and percutaneous strategy.

CASE REPORT

A 32-year-old female patient presented with pain in the right hand and forearm that was exacerbated with movement. Her medical history revealed an acute right axillary arterial occlusion 4 years earlier treated with a surgical embolectomy. A work-up to assess the

Özet– Arteriyel torasik çıkış sendromu, subklavyen veya aksiller arterin kostalar ve skalen kasları arasında sıkışmasından kaynaklanır. Hastalar çeşitli klinik durumlarla ortaya çıkabilir. Bu yazıda, periferik emboli ile başvuran bir arteriyel torasik çıkış sendromu olgusu sunulmaktadır. Arteriyel torasik çıkış sendromu nadir görülen periferik emboli nedenidir. Tanı yaklaşımı ve tedavi yöntemleri bu olgu sunumunda örnek hasta üzerinden tartışılmıştır. Klinisyenler bu tanıyı akılda tutmalı ve klinik durumuna göre yönetmelidir.

cause of the thromboembolism had been unremarkable, with the exception of a homozygous methylenetetrahydrofolate reductase (MTHFR) polymorphism. Vascular anatomical imaging had not been performed during or after the thromboembolism attack. Her follow-up treatment had been acetylsalicylic acid and folic acid. The current symptoms started 1 day prior to presentation while holding her 1.5-year-old child on her lap. A physical examination revealed coolness and a loss of radial pulse suggestive of acute limb ischemia.

A Doppler ultrasound examination revealed diminished blood flow in the right upper extremity arteries. A heavy thrombus burden in the right upper extremity arteries (Fig. 1a, Video 1*) and a cervical rib with a subclavian arterial aneurysm (Fig. 1b, Video 2*) suggestive of TOS were detected on peripheral angiogra-

Abbreviations:

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| TPA | Tissue plasminogen activator |
| TOS | Thoracic outlet syndrome |

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Correspondence: Dr. Berhan Keskin, Kartal Koşuyolu Yüksek İhtisas Eğitim ve Araştırma Hastanesi, Kardiyoloji Kliniği, İstanbul, Turkey.

Tel: +90 216 - 500 15 00 e-mail: bekeskin@ku.edu.tr

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phy. The presence of axillary arterial collateralization and a brachial thrombus led to the opinion that there was both acute and chronic thrombosis. Anticoagulation with intravenous unfractionated heparin was initiated. Computed tomography imaging confirmed compression of the subclavian artery and the diagnosis of TOS (Fig. 1c, Video 3*).

The management of intra-arterial thrombolysis has been similar to that of lower extremity acute ischemia; however, there is the risk that an unstable catheter position may cause inadvertent subclavian aneurysm trauma, carotis, or vertebral artery injury. The possibility of an unintended positional change of the catheter may necessitate tissue plasminogen activator (tPA) administration to the carotid or vertebral artery. For all of these reasons, in this case, intravenous, slow-infusion, low-dose tPA (alteplase, 1 mg per hour for 24 hours) was initiated. The extremity

coolness and resting pain diminished after the tPA infusion; however, pain with movement continued. The infusion of unfractionated heparin was maintained for 48 hours due to the persistence of symptoms with movement. After completion of the thrombolytic therapy, a control angiography revealed a decrease in the thrombus burden; however, the axillary arterial collateralization and diminished flow was compatible with a chronic component (Video 4*). A decision was made to pursue interventional therapy, and balloon angioplasty was performed in the narrowed segments with 5*100-mm and 6*100-mm balloons with a 0.035" balloon dilatation catheter (Mustang; Boston Scientific, Marlborough, MA, USA). After the procedure, the patient's arterial flow improved and her symptoms resolved (Fig. 1d, Video 5*). A cervical and first rib resection along with vascular reconstruction was planned, but the patient declined the surgery. She was

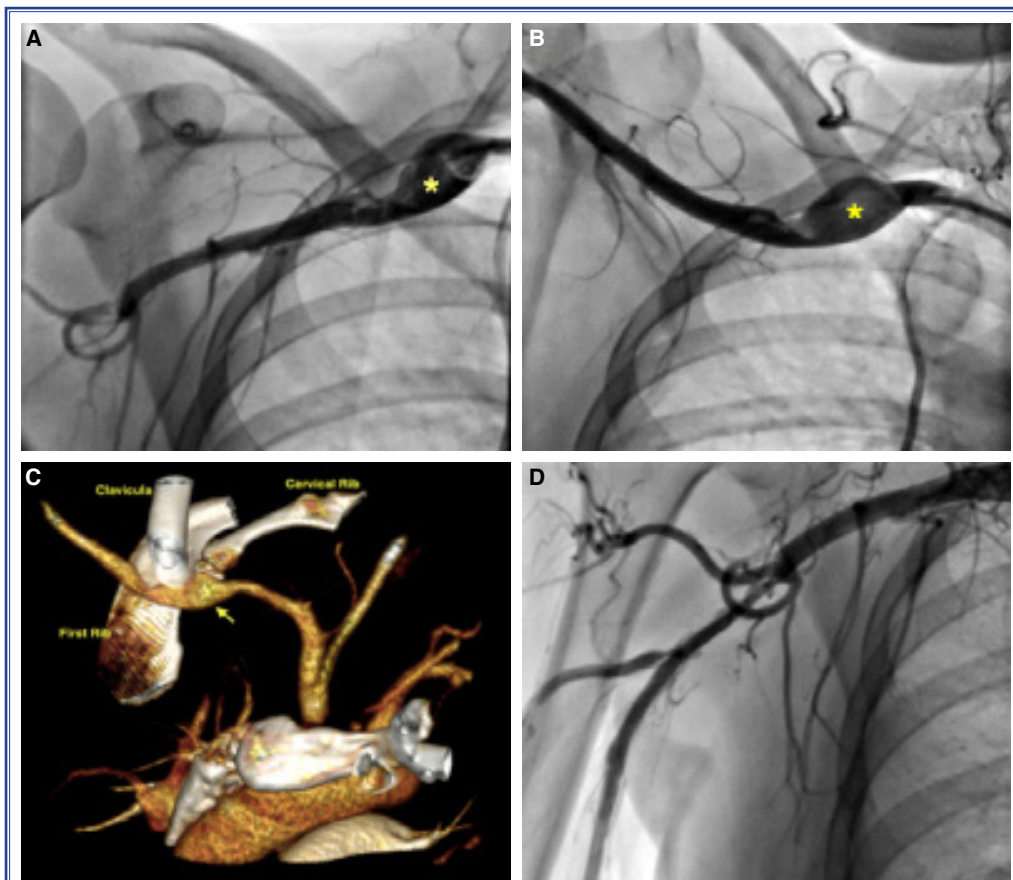


Figure 1. (A) Selective angiography revealed occlusion of the axillary artery, a thrombosed subclavian artery aneurysm, and a brachial arterial thrombus; (B) A selective angiography image of the subclavian artery aneurysm; (C) A 3-dimensional computed tomography image illustrating cervical rib compression of the aneurysmal subclavian artery; (D) Image of improved arterial flow in the axillary artery and distal arterial bed after balloon angioplasty.

prescribed oral anticoagulation medication of apixaban 5 mg twice daily and was well at a 6 month follow-up.

DISCUSSION

Arterial TOS is an uncommon entity in daily practice, but one that should be remembered and clinicians must be able to recognize the findings.^[1] It is generally asymptomatic, but symptoms may be aggravated by repetitive trauma or ischemic events.^[1,3] Arterial TOS can also lead to the formation of a subclavian artery aneurysm or be complicated by chronic or acute ischemia.^[1] A thorough history, physical examination, and imaging are crucial for the diagnosis. Doppler ultrasound and angiography can be used to evaluate blood flow in the extremities. Angiography and computed tomography can demonstrate arterial compression or the presence of a cervical rib or subclavian artery aneurysm. Pharmacological, interventional, or surgical methods may be preferred, according to the clinical scenario.^[4]

Cardiologists and cardiovascular surgeons should keep arterial TOS in mind particularly in cases of a young patient with an undetermined source of upper limb arterial embolism (i.e., those with sinus rhythm, absence of intracardiac thrombus, mass or patent foramen ovale, absence of atrial fibrillation in rhythm monitoring).

In patients with chronic symptoms, surgical decompression and vascular reconstruction is generally the first treatment option.^[4] In patients with acute thromboembolism and limb-threatening ischemia, urgent treatment should be considered. Current approaches include surgical embolectomy, balloon angioplasty, thrombolytic therapy, and anticoagulation.^[5] In the present case, acute limb ischemia was successfully managed with percutaneous intervention and fibrinolytic treatment. A graft-stent implantation was not considered in the absence of rib resection due to a high risk of stent fracture, thrombosis, and restenosis.^[4]

Arterial TOS is infrequent and can present

with different clinical appearances. Patients may be asymptomatic or present with acute or chronic limb ischemia symptoms. A cervical rib, subclavian aneurysm, and blood clots are clues to suspect arterial TOS. Cardiologists and cardiovascular surgeons should be cognizant of arterial TOS in patients with an undetermined source of upper limb thromboembolism. Multimodality imaging is required to confirm the diagnosis. If the patient refuses surgery, thrombolysis with low-dose, slow-infusion tPA followed by balloon angioplasty can be an acceptable approach.

*Supplementary video file associated with this article can be found in the online version of the journal.

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REFERENCES

1. Grunebach H, Arnold MW, Lum YW. Thoracic outlet syndrome. *Vasc Med* 2015;20:493–5. [[CrossRef](#)]
2. Criado E, Berguer R, Greenfield L. The spectrum of arterial compression at the thoracic outlet. *J Vasc Surg* 2010;52:406–11. [[CrossRef](#)]
3. McCarthy WJ, Yao JS, Schafer MF, Nuber G, Flinn WR, Blackburn D, et al. Upper extremity arterial injury in athletes. *J Vasc Surg* 1989;9:317–27. [[CrossRef](#)]
4. Hussain MA, Aljabri B, Al-Omran M. Vascular Thoracic Outlet Syndrome. *Semin Thorac Cardiovasc Surg* 2016;28:151–7.
5. Marine L, Valdes F, Mertens R, Kramer A, Bergoing M, Urbina J. Arterial Thoracic Outlet Syndrome: A 32-year Experience. *Ann Vasc Surg* 2013;27:1007–13. [[CrossRef](#)]

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