

## Critical ischemia of the left arm caused by thoracic outlet syndrome (TOS): case report and review of the literature

TO THE EDITOR: A 31-year-old woman came to our attention for critical ischemia of her left arm, with pain, numbness and functional impotence, especially when abducting the shoulder. The patient had previously been visited at another hospital, with an indication for medical therapy: ASA 100 mg daily, cilostazol 50 mg twice a day, pentoxifylline 600 mg twice a day.

Her past medical history included two pregnancies and estro-progestinic therapy for contraception. She was an active smoker (20 cigarettes per day). Apart from the presence of an anxiety syndrome, the patient reported having an active life, with good tolerance to exercise.

At clinical examination, her left hand was hypothermic and pale, with cyanosis of the fingers, nail dystrophy and pulselessness of brachial, ulnar and radial artery. In the left supraclavicular fossa a bony protuberance was palpable above the clavicle.

A Duplex ultrasound showed occlusion of the ipsilateral subclavian-axillo-humeral arterial axis

caused by a cervical rib. Furthermore, there was a left thrombosed supraclavicular aneurysm of the subclavian and axillary artery. A small calibre ulnar artery was patent for its first 5 cm, with a poor recanalization signal. The radial artery was absent, as well as the palmar arch.

The patient was, therefore, admitted to our division, and underwent a CT-angiography (Figure 1) and a selective angiography of the upper left limb with static and dynamic poses (Figure 2); both confirmed Duplex ultrasound findings.

Having obtained an informed consent from the patient, she underwent surgery under general anesthesia for removal of cervical left rib, followed by an unsuccessful attempt of thromboembolectomy with Fogarty catheter of the humeral arterial axis. Therefore, to correct the arm ischemia, a subclavian-humeral bypass was made using a reversed great saphenous vein taken from her right thigh. We also performed an endarterectomy of the distal brachial artery and a surgical angioplasty of the passage be-

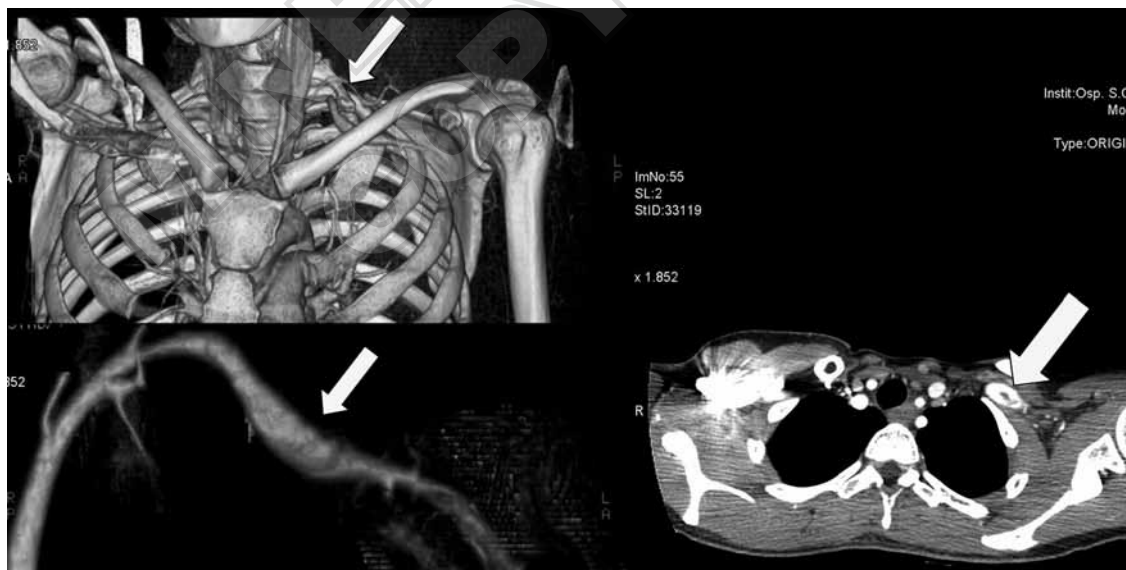


Figure 1.—Preoperative CT-scan. Note left cervical rib and the subclavian artery aneurysm.



Figure 2.—Preoperative angiography with static and dynamic poses. In last images note the poor recanalization of vessels at the forearm and absence of perfusion at left hand.

tween brachial and ulnar artery with a bovine pericardium patch. At the end of the procedure humeral and interosseous arteries showed good pulsatility and there was a good perfusion in both proximal and distal ulnar artery and the hand was adequately perfused.

The postoperative course was complicated by the presence of a small pneumothorax in the left apical region, associated with subcutaneous emphysema of the left lateral chest wall, which were caused by a small pleural gap opened while removing the cervical rib. The patient however remained asymptomatic from the respiratory point of view.

The patient was also visited by the hematologist to assess the possible thrombophilic diathesis: a mutation of MTHFR gene in a heterozygous genotype was found; oral anticoagulant therapy was indicated for at least six months.

The patient was discharged on the 14<sup>th</sup> postoperative day: her left upper limb was warm, pink and well perfused down to the fingers; both ulnar and brachial pulses were palpable. A Duplex ultrasound before discharge revealed the patency of the bypass with good flows up to palmar arch. An angio-CT scan confirmed the patency of the bypass and the regular success of the surgical resection of cervical rib (Figure 3).



Figure 3.—Postoperative CT scan. Patency of bypass and removal of the cervical rib. Reworking of CT images using OsiriX®.

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Four months later the patient has completely recovered her left shoulder, arm, forearm and hand function. Both radial and ulnar pulses are palpable.

The thoracic outlet syndrome (TOS) includes a number of clinical conditions determined by the compression of the neurovascular bundle due to an anatomical or functional abnormalities of the thoracic outlet. The presence of a cervical rib is the most frequent anomaly, affecting 0.5-1% of the general population.<sup>1</sup>

In 95% of cases, TOS presents with symptoms exclusively or predominantly neurologic (n-TOS); vascular involvement is estimated at 5%, being 3% venous and 1-2% arterial.<sup>2</sup>

Arterial complications are rare but dangerous, with clinical features ranging from the development of an aneurysm of the subclavian district to an acute or critical ischemia that may threaten the viability of the upper limb, very dramatic events which could hesitate in irreversible ischemia.<sup>3</sup>

The clinical features are variable and may be due to the different degree of compression of vessels and nerves, which can lead to complications. Symptoms are frequently mixed.<sup>4</sup>

The extremely high proportion of nervous involvement, as well as the difficulty in making correct diagnosis of TOS,<sup>3</sup> may lead to the underestimation of vessels involvement.

A critical ischemia that threatens the viability of the upper limb can therefore develop, as a result of chronic damage to the intimal wall of the vessel.<sup>4</sup> Repeated trauma in fact can trigger the development of an aneurysm and its thrombosis. The latter mechanism can compromise the peripheral arterial circulation with repeated embolization, making revascularization more challenging.

In the presence of neurological symptoms of TOS, it is therefore mandatory to rule out a possible arterial involvement. Differences between radial pulses and systolic blood pressure between the two arms must be investigated. If these findings are normal at rest, any changes in the radial pulse during the execution of appropriate manoeuvres to activate the shoulder girdle should be sought. The diagnosis of TOS however remains one of the most difficult to formulate.<sup>2</sup> Moreover, as emerging from a careful review of the literature, widely accepted standards for the diagnosis are lacking.

As for its therapy, TOS treatment has been a major topic of discussion for several decades. A review of the literature suggests that conservative treatment is considered the first choice. It seems to be more effective than surgery in terms of recovery of work, although its superiority compared to non treatment or placebo effect has not been demonstrated yet.<sup>5</sup>

Moreover, there is not currently enough evidence that the established interventions for thoracic outlet syndrome are helpful in relieving pain.

However, in presence of neurological or vascular complications, or after failing of a rehabilitative therapy performed for 3 to 6 months, surgical treatment is generally required. Arterial aneurysms and occlusions in the presence of a permanent or postural arterial compression should be treated as soon as possible, because of their tendency to cause embolization, threatening limb viability.<sup>5</sup>

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