Arterial complications of thoracic outlet syndrome

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ABSTRACT

The clinical manifestations of thoracic outlet syndrome are mainly neurological. Although arterial complications are rare, they are potentially severe. Among these are aneurysms associated with embolism and thrombosis. The authors report a case of a 37 year-old woman with bilateral cervical rib that developed embolism in the right upper limb from a poststenotic right subclavian artery aneurysm and dilatation of the left subclavian artery, both due to compression.

Keywords: Thoracic outlet syndrome, cervical rib, arterial complications.

RESUMO

As manifestações clínicas da síndrome do desfiladeiro torácico são predominantemente neurológicas, sendo as complicações arteriais raras, mas potencialmente graves. Entre elas, devemos citar os aneurismas com complicações embólicas e a trombose. Os autores relatam o caso de uma mulher de 37 anos com costela cervical bilateral que apresentou embolia no membro superior direito originada de um aneurisma pós-estenótico da artéria subclávia direita, além de apresentar ectasia da subclávia esquerda também por compressão.

Palavras-chave: Síndrome do desfiladeiro torácico, costela cervical, complicações arteriais.

Introduction

The expression "thoracic outlet syndrome" (TOS) was first used by Peet[1] to describe patients with neurovascular symptoms relative to possible sites of compression of the neurovascular bundle, which are basically three: interscalene triangle, costoclavicular space and retro-pectoralis minor space.

It is believed that the combination of two factors is required to cause this syndrome: 1) anatomical narrowing; 2) some type of trauma that triggers symptoms.[2]

Compression in the interscalene triangle, known as scalene syndrome, has neurological and arterial symptoms, there are no venous symptoms, since the subclavian vein is not contained in this triangle. Costoclavicular compression syndrome can compress any bundle structure. Compression in the retro-pectoralis space, or small pectoral syndrome, is rare, and treatment is essentially clinical.

There are other possible compression sites,[3] with similar manifestations. Among them we can cite the median syndrome, in which there is compression of the axillary artery between brachial plexus bundles, and Langer's syndrome, in which there is compression of the vasculonervous bundle by an anomalous muscle, the axillopectoral muscle.

The main symptoms are pain and parestesias, which occur spontaneously or are caused or exacerbated by movements that reduce the dimensions of potential compressions spaces. Its distribution is more frequent in the volar aspect of the forearm and of the last two fingers.

Initial historic publications described almost exclusively vascular manifestations.[4] Throughout time, more knowledge of this syndrome was acquired; it has been observed that neurological symptoms were much more frequent, and currently it is known that they account for most cases, corresponding to approximately 95% of clinical complaints. Presence of changes in arterial pulses, such as maneuvers that simulate compression, do not necessarily indicate that there is arterial lesion.

Arterial complications are rare, but potentially more severe than neurological manifestations, and may lead to significant sequelae.

The authors report the case of a patient with acute arterial occlusion in the right upper limb due to embolism secondary to a poststenotic aneurysm as TOS complication.

Case report

A 36-year-old female patient presented with complaint of cyanosis, pallor and parestesias in the right upper limb for 3 days. She denied complaint of previous parestesia in the upper limbs. She had no comorbidity, was not a smoker and was not using oral contraceptives.

On initial physical examination, the patient had finger pallor and right hand with reactive hyperemia. She had no radial and ulnar pulses in the right upper limb, normal pulses in the contralateral limb and in the lower limbs. Adson's maneuvers performed bilaterally had positive results.

Apical lordotic chest x-ray showed complete and articulated bilateral cervical rib and articulated in the first rib bilaterally (Figure 1).

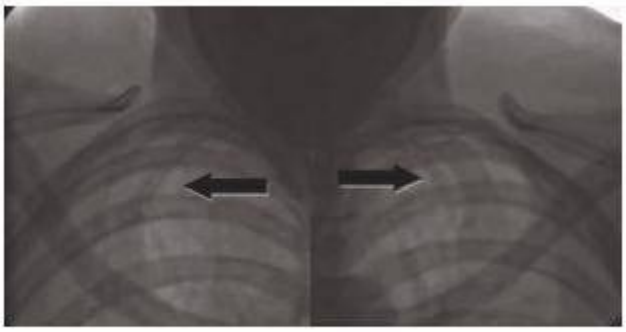

Figure 1 - Apical lordotic chest x-ray (arrows indicate bilateral complete cervical rib)

Doppler ultrasound of the upper limbs was performed, showing occlusion of the radial and ulnar arteries in the proximal third of the right forearm. It also showed positivity for arterial compression tests through scalene maneuvers, with expressive reduction in peak systolic velocity in the axillary artery.

Digital angiography through femoral puncture showed right subclavian artery aneurysm, with embolic occlusion of forearm arteries, and left subclavian artery ectasia (Figure 2).

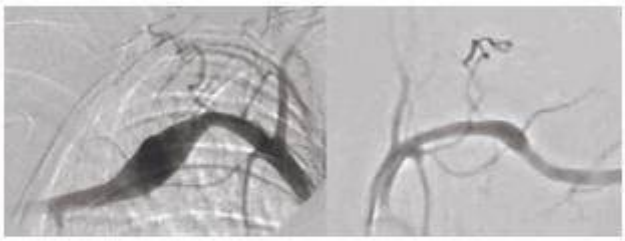

Figure 2 - Arteriography of supraaortic trunks showing right subclavian artery aneurysm and left subclavian artery ectasia

The patient was submitted to surgery using a right anterior cervicotomy approach. The right cervical rib was resected, using anterior scalenectomy and excision of the right subclavian artery aneurysm (Figure 3) with interposed great saphenous vein graft. The patient's course was satisfactory and uneventful in the postoperative period.

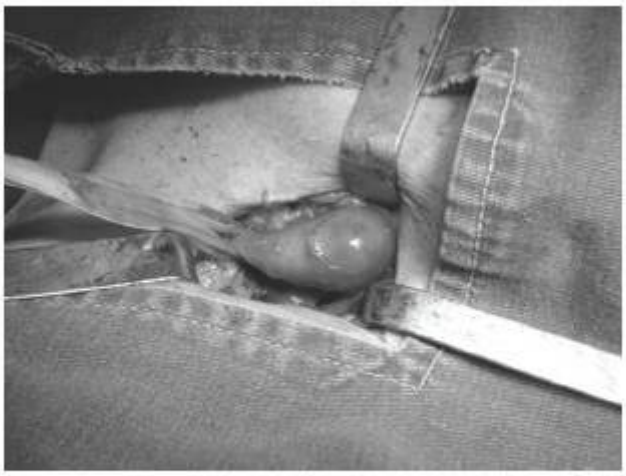

Figure 3 - Intraoperative image of the right subclavian artery aneurysm

Thirty days later, she was submitted to resection of the left cervical rib using anterior scalenectomy. She had good evolution and no complications.

Discussion

Cervical ribs and the first anomalous rib are rare conditions, present in approximately 1% of the population and in 4.5% of patients with TOS.[5] They are usually findings of imaging examinations, such as chest x-rays. Most are completely asymptomatic, and rarely require treatment. Cervical ribs and the first anomalous rib may have complications that are potential threats to the upper limb, requiring regular follow-up.

According to Sanders et al.,[5] symptom onset is preceded by cervical trauma in 80% of cases, and 20% of cases occur spontaneously. However, in a subgroup of 12 patients who had complete cervical rib, the incidence of spontaneous symptoms reached 50%. Of a total of 40 patients with cervical rib, it measured 1-2 cm in 11 cases; 2-5 cm in 17 cases; and were complete in the other 12 patients. In cases of complete rib, three were articulated directly with the first rib, and the others were inserted in the last rib through a fibrous ligament.

The type of cervical rib is of great significance in arterial complications. It has been well established, after a study by Gruber,[6] that short (type I) and incomplete ribs (type II) preferentially produce neurological complications, while long or complete ribs (type III) have arterial complications.[6] Sanders et al.[7] confirmed the same findings. Short et al.[4] showed that 75% of patients with incomplete cervical rib had their symptom onset associated with cervical trauma, while in patients with complete cervical rib only 50% of cases had this association.

Our patient had complete bilateral cervical rib (type III), with arterial complications, right subclavian artery aneurysm and left subclavian artery ectasia. The most severe dilatation to the right was possible due to the fact that the patient is right-handed, since most studies attempt to correlate TOS with upper limb trauma.[8]

Clinical manifestations comprehend a wide range of symptoms, and not all are necessarily present in the clinical status of each patient. Most symptoms are of neurological compression, and these are manifested as pain in the cervical region, shoulder, anterior thorax, elbow, arm, forearm, and

occipital headache. Reports of finger parestesias are common, more frequently in the fourth and fifth fingers, due to compression of lower roots of the ulnar nerve, reduction in hand and arm strength, and there is usually symptom worsening with limb elevation. There may also be reduced temperature, Raynaud's phenomenon and increased hand sweating. The incidence of each symptom is similar to that of patients who have no cervical rib.

Arterial lesions in TOS are usually due to bone abnormalities. Findings in the literature correlate with presence of cervical rib in 70-100% of cases. In these patients the subclavian artery passes over the cervical rib (which compresses it in its inferior aspect) and produces an intimal lesion with or without poststenotic dilatation.[9] Such patients often have embolic complications in their course, which are the most disabling aspect of this disease and its treatment is difficult. Arterial complications tend to be treated late in relation to their neurological equivalents. In fact, most of the times the basal problem is not recognized until the thromboembolism has occurred, whether due to an intimal lesion of the subclavicular segment or due to poststenotic dilatation or aneurysm.[10]

Presence of anomalous first rib and muscle changes increases probability of arterial lesion and embolic phenomena. The first anomalous cervical rib leads to changes similar to those of the cervical rib, but it affects the artery in its upper aspect. Anterior scalene muscle hypertrophy in athletes is associated with subclavian artery impairment, and its occlusion with distal embolism.[10]

The need of early detection of lesions caused by emboli at an early stage is extremely important, and all patients with first rib abnormalities or with cervical rib should be regularly followed using Doppler ultrasound of the subclavian-axillary segment.[10] Every stenotic segment or presence of aneurysm should be later assessed by angiography to program a surgical intervention.

Many patients with severe ischemia who seek for medical assistance have previous history of warning episodes. Patients with history of unilateral Raynaud's phenomenon (especially in the radial territory)[11] and patients experiencing sudden pain in the digital pulp with vasomotor changes (pallor or cyanosis) should be suspected of having an embolic event and Doppler ultrasound and angiography of the thoracic outlet should be performed.[12]

Chest and cervical spine x-rays are extremely important to reveal bone changes. Computed tomography can be useful in the TOS diagnosis, especially in patients in whom x-rays are normal.[13] Magnetic resonance has proved to be efficient in showing fibrotic bands and deviation of the brachial plexus in patients without cervical rib.[14] Electrodiagnostic assessment is useful for the diagnosis of compression in the carpus and Guyon's canal, but it is altered for the diagnosis of TOS due to the position in which this test is performed.[15,16] Komanetsky et al., in a study of 21 patients with TOS and 23 controls, in a relaxed and forced position, did not consider this test useful for diagnosis.[17]

The treatment of this disease can be conserved through physical therapy, rest of the activity that caused the symptoms and analgesia when the syndrome is mainly of neurological impairment.[1] Surgical treatment should be indicated in cases of failure in clinical treatment (after 6 months without improvement or symptom recurrence),[1] uncontrolled pain and parestesia, in addition to vascular impairment (pallor, reduced pulses, cyanosis, Raynaud's phenomenon,[11] limb claudication or pain at rest or ulcers).[1]

There are two surgical techniques for correction: supraclavicular and transaxillary approach.[5,7,18] The first consists of an approach that can be infra- or supraclavicular with platysma incision, anterior and medial scalenectomy, besides resection of the first rib exposing the brachial plexus and the subclavian artery and vein.[5,7,18,19] The second uses an approach through the ipsilateral axilla, resecting the first rib but preserving the scalene muscle.[19,20]

Studies have shown that success rates have been similar between both techniques,[5,7] although the supraclavicular approach allows for a better visualization of the scalene muscle. In addition. when the scalene muscle has any abnormality that could be responsible for the syndrome, it is possible to incise it at this moment; on the other hand, using the transaxillary approach, such procedure would be performed at a different surgical time.

One of the main complications of the surgery has been the phrenic nerve lesion,[5] which usually regressed after 3 months in most cases. Other complications that have been reported are subclavian artery and vein lesions. The first is much more frequent than the second.[5,7,19,20] Pneumothorax has also been reported. It is more frequent when the transaxillary approach is used,[7,19,21] but thoracocentesis was rarely required.

As the patient of the present report had a vascular TOS (pure arterial impairment) due to compression of a real cervical rib, a supraclavicular approach was used with resection of the cervical rib associated with right scalenectomy. Furthermore, aneurysm resection was performed using an interposed saphenous vein graft. That approach was adopted due to the unrestricted exposure of the interscalene triangle and good access for cervical rib resection.[7,12] The same technique was used for cervical rib resection on the left side, with no need of arterial resection, since the subclavian artery in that side was only dilated.

TOS is a common disease that has complex anatomy and a large anatomical variation of structures, which should be managed by a specialized team and vascular surgeon experienced in this type of surgery.

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