Clinical Case Report

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# Thoracic outlet syndrome in a postoperative cervical spondylotic myelopathy patient

## A case report

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**Abstract**

**Rationale:** Thoracic outlet syndrome (TOS) is a rare disease that presents with neurogenic and vascular symptoms similar to those of cervical spondylosis. However, making the diagnosis of TOS can be challenging due to a lack of standardized objective confirmatory tests.

**Patient concerns:** A 66-year-old man presented with neck, supraclavicular, and right shoulder pain as well as numbness and weakness in the right arm after surgery to correct cervical spondylotic myelopathy (CSM).

**Diagnosis:** Magnetic resonance imaging confirmed the diagnosis of CSM. He was diagnosed with TOS based on the manifestations and examination findings.

**Interventions:** After surgery for CSM, nonoperative management was provided.

**Outcomes:** The patient reported pain relief and improving sensation in the shoulder and supraclavicular region.

**Lessons:** Based on this case and the reviewed literature, to optimize the diagnosis and treatment of CSM, clinicians should consider preoperative differential diagnosis to preliminarily exclude it.

**Abbreviations:** ATOS = arterial thoracic outlet syndrome, CSM = cervical spondylotic myelopathy, CT = computed tomography, CTA = computed tomography angiography, MRA = magnetic resonance angiography, MRI = magnetic resonance imaging, NTOS = neurological thoracic outlet syndrome, TOS = thoracic outlet syndrome, VTOS = venous thoracic outlet syndrome.

**Keywords:** cervical spondylotic myelopathy, diagnosis, postoperative, thoracic outlet syndrome

## 1. Introduction

Thoracic outlet syndrome (TOS) is a rare condition caused by the extrinsic compression of neurovascular structures passing from the upper thoracic aperture to the axilla through the thoracic outlet. The thoracic outlet is the cervicoaxillary canal defined as the channel from the supraclavicular fossa to the axilla that passes between the clavicle and the first rib.[1,2] Subjects with neurovascular bundle impingement at the thoracic outlet have been gathered together under the name of TOS. The definition of TOS, although controversial, is generally accepted as "upper-extremity symptoms due to compression of the neurovascular bundle in the area of the neck just above the first rib."[3]

TOS can be caused by compression of the nerves of the brachial plexus, the subclavian vein, and/or the subclavian artery at several sites between the first rib and the clavicle.[4] Patients can be categorized into having neurological (NTOS), venous (VTOS), and/or arterial (ATOS) according to the predominant clinical presentation.[5,6] NTOS is the most common subtype, accounting for over 90% of all TOS cases.[3] Patients with NTOS often present with pain, paresthesia, weakness, or numbness in the upper limbs. Symptoms of VTOS include acute pain, arm swelling, and blue discoloration. ATOS accounts for less than 5% of all TOS cases. Affected patients often experience arm pain, weakness, coolness, pallor, and paresthesia.[5,7,8]

Making the diagnosis of TOS can be challenging, as criteria are controversial, and currently, there are no confirmatory tests, which commonly lead to delayed diagnosis in clinical practice. Evaluations are made only if the compression is severe.[9] A meticulous physical examination of potential TOS patients is a basic step in establishing a TOS diagnosis, followed by the collection of evidence related to history and clinical manifestations. Maneuvers to evaluate peripheral pulses and pressures by neurologic testing using compressive and provocative tests, such as the Adson, Wright, and Roos tests, as well as hyperabduction, can support the diagnosis.[10–13]

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Most cases are thought to result from an anatomic predisposition with superimposed neck trauma from either a single acute incident or repetitive stress.[2,14] TOS is also reportedly caused by a variety of diseases, and reports have suggested that examiners differentiate patients with TOS from those whose symptoms are related to brachial plexus compression that is not related to thoracic outlet pathology, such as cervical spondylosis, shoulder dysfunction, and other compression neuropathies.[2] Limited case reports have associated TOS with postoperative cervical spondylotic myelopathy (CSM). Here, we describe the case of a postoperative CSM patient with TOS.

## 2. Case report

A 66-year-old man presented with a 15-year history of neck, supraclavicular, and right shoulder pain, numbness, and weakness in the right arm. He was previously diagnosed with CSM but hesitated to undergo surgery until the symptoms worsened suddenly 5 years later. He underwent C3–6 posterior decompression with laminoplasty and internal fixation (Fig. 1A and B), followed by lumbar dynamic transpedicle fixation for lumbar disc herniation 6 months later. However, the surgical outcome was not very satisfactory: He was not able to walk without assistance and his arm weakness and numbness improved little. A physical examination revealed muscle weakness (Lovett grade 2/5) of the intrinsic muscles of the right hand accompanied by decreased grasp strength. Significantly reduced sensation within the ulnar part of the forearm was noted. He had a positive Adson test result and minor muscle atrophy of the thenar muscles. His arm abduction was limited to 30°, which prevented conduction of the full Wright and Roos tests. In the Adson test posture, the patient had slight C-8 radiculopathy; electrodiagnostic studies demonstrated lower trunk axonal loss. However, plain x-ray, inpatient magnetic resonance imaging (MRI), and three-dimensional CT of the neck failed to reveal the cervical rib or prolongation of the transverse process of the seventh cervical vertebra. The patient refused surgery as a first-choice intervention since he had already undergone 2 surgeries. Thus, he accepted nonsurgical management such as physical therapy (e.g., muscle stretching, muscle strengthening, range of motion exercises) and acupuncture. At the 6-month and 1-year follow-up points, the patient reported improvements in his right hand despite numbness in the digits. He reported pain relief in his shoulder and supraclavicular area. The Adson test result was negative and an examination revealed improved sensation in the ulnar part of the arm. His intrinsic hand strength had improved to Lovett grade 4/5. The Medical Ethics Committee of Affiliated

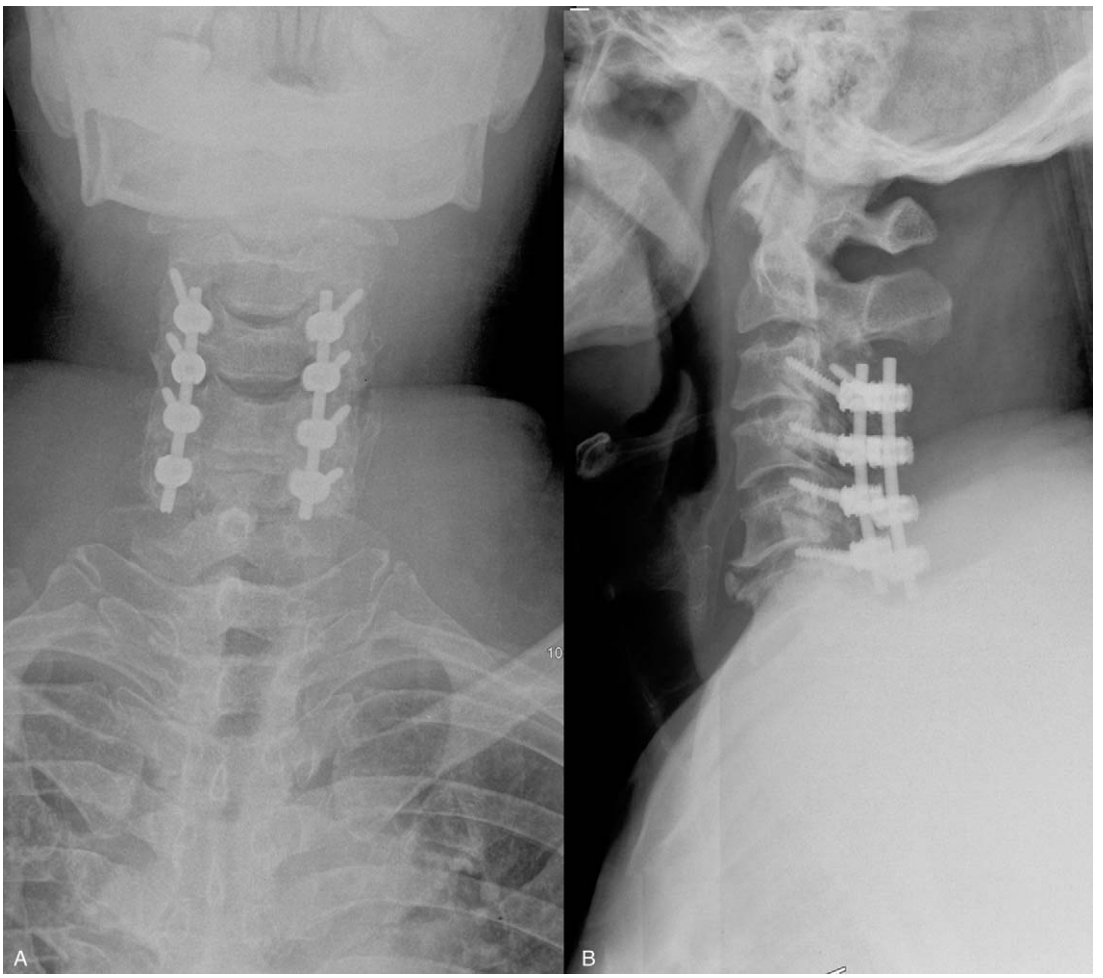


**Figure 1.** Postoperative radiography of the neck shows the surgery of C3–6 decompression with laminoplasty and internal fixation (A, anteroposterior view; B, lateral view).

Longhua Hospital of Shanghai University of Traditional Chinese Medicine approved this clinical experiment, and the patient provided informed consent for the publication of this case report and any related images.

## 3. Discussion

TOS is a well-described disorder that results from thoracic outlet compression of the brachial plexus and/or subclavian vessels. TOS generally occurs in young patients with a greater prevalence in women aged 20 to 40 years.[10] The etiology of TOS is complex and not completely understood. It can be attributed to bony-tissue anomalies such as first-rib abnormalities, cervical ribs, and bony tubercles. Soft-tissue anomalies such as fibrous bands, scalene muscles, and postural problems also play an important role.[10,15]

The diagnosis of TOS remains challenging due to its varied clinical manifestations and lack of confirmatory testing and generally accepted diagnostic criteria.[2,15] As comprehensive knowledge of TOS has increased, various methods have recently been applied to evaluate it. Cervical spine or chest radiographs can help with the identification of cervical ribs, prominent C7 transverse processes, and other osseous abnormalities that may cause compression at the thoracic outlet. Three-dimensional CT and MRI can also assist with evaluating for congenital anomalies and space-occupying diseases; however, limited studies to date have examined this issue and axial slices of interesting regions could miss such anomalies.[16] Moreover, TOS is usually dynamically caused by compression of the branchial plexus by a taut fibrous band, which is not usually clearly presented on MRI or CT scans.[11] CT angiography (CTA) and magnetic resonance angiography (MRA), particularly the latter, are currently accepted as accurate methods for investigating VTOS; however, the role of this technology during diagnosis remains to be elucidated.[2,6] Duplex ultrasonography scanning is considered a helpful workup approach, but the results often depend on operator experience.[6,17] Findings of neurophysiologic studies used to be regarded pathological in TOS cases only if the nerve injury was severe.[2] However, it turned out that nerve fibers may undergo changes, manifesting with decreased nerve conduction velocity.[18] To our knowledge, the evaluation of potential TOS primarily depends on a meticulous physical examination, clinical presentation, detailed history, abnormal electrophysiological test results, and positive provocative test results. The purpose of further imaging is to confirm the diagnosis and classification of TOS. Since MRA and CTA are not cost effective and our patient was averse to contrast agent use, we did not perform them. A novel electrophysiological study demonstrated the usefulness of the triple stimulation technique and recommended other methods such as motor-root stimulation. Since the electrophysiological diagnosis of TOS is difficult, advances in the field are of great clinical significance since they may assist with diagnostic localization. We look forward to further electrodiagnostic studies.

Surgical and nonsurgical treatments are common interventions for TOS. Surgical interventions for TOS include resection of the first rib or cervical rib, scalenectomy, scalenotomy, and the division of fibrous bands.[15] There are 3 main surgical approaches: transaxillary, supraclavicular, and posterior subscapular. In practice, they all depend on surgeons, who may combine the interventions or make other choices, such as bypass surgery.[2,19] However, most patients are inclined to choose nonsurgical interventions first,[15] which aim to decrease the compression of the affected areas. Conservative management consists of education, lifestyle modifications, physical therapy, and medications. Published studies that treated patients with rehabilitation programs, home exercises, heat pack therapy, cervical traction, stretching, and range of motion exercises also count.[15,20]

Decompression of the brachial plexus is a technically demanding surgical procedure that requires expertise in peripheral nerve, vascular, and thoracic surgery; moreover, it carries the potential risk of reoperation in unresolved recurrent cases.[4,9] In cases of reoperation, the TOS surgery is more challenging from a diagnostic and technical standpoint.[21] The patient in our case refused surgery and chose nonsurgical treatment. Thus, we taught him muscle stretching, muscle strengthening, and range of motion exercises that he could perform at home. He completed several acupuncture sessions to release the adhesion. At the 1-year follow-up mark, he reported pain relief in his shoulder and supraclavicular areas. His Adson test result was negative and the sensation within the ulnar arm had improved. His intrinsic hand strength had improved to Lovett grade 4/5. However, the numbness in the digits and the pinch strength had improved marginally, so we reported his case at the 15th Annual Conference of the Cervical Spondylosis Subcommission of the Chinese Association of Rehabilitation Medicine to solicit the opinions of experts, who considered that the numbness should partly be attributed to an unsatisfactory CSM surgical outcome.

Our case has several limitations. First, we did not perform complete Wright or Roos tests due to the patient's low degree of shoulder abduction; nevertheless, the specificity of the Adson test was 79%.[22] Second, we did not perform CTA or MRA imaging due to patient refusal; thus, the precise compression location remained unknown. The clinical manifestations and detailed history of our patient were strongly consistent with a diagnosis of TOS. However, the lack of a further diagnosis to ascertain the compression site resulted in a final diagnosis of nonspecific TOS,[23,24] but given the complexity of diagnosis and the overlapping of the patient's symptoms with those of other diseases, an accurate diagnosis of TOS can be a substantial challenge in practice.[25]

Studies tend to exclude cervical problems to allow for a precise diagnosis of TOS. To our knowledge, few studies have confirmed the diagnosis of TOS in postoperative patients with cervical disc disease and spondylosis, but all patients underwent anterior cervical discectomy and fusion and had bony anatomic abnormalities.[12] Here, we report a TOS case without bony abnormalities treated with spinal posterior approach surgery since an anterior approach may cause adhesions of the ligaments or bands due to long-term intraoperative traction of the scalene musculature. Although the specific compression location is unknown, according to literature reports, over 70% of TOS cases are caused by soft-tissue anomalies. After confirming the lack of congenital anomalous bony findings, we ultimately treated the patient with conservative interventions.

However, we had no information about the patient's clinical presentation before surgery; thus, we are unsure about whether the TOS is secondary to the CSM surgery. Orthopedists who do not regularly treat patients with TOS may not have an accurate view of this disorder and may not be aware that it may occur in CSM patients. Here, we highlight the importance of differentiating TOS from cervical diseases. A preoperative TOS differential diagnosis can be made in CSM patients to ensure accurate diagnosis and treatment. Whether CSM patients with TOS, especially those with hypertrophy, can undergo combined surgery should be further studied. Whether postoperative CSM

patients have unsatisfactory outcomes due to insidious TOS requires further studies in the future.

## 4. Conclusions

TOS may occur in CSM patients. Here, we highlight the importance of differentiating TOS from cervical diseases since most orthopedists are unaware of this syndrome. The accurate differentiation of TOS from cervical diseases in postoperative CSM patients may allow clinicians to make optimized treatment decisions. Furthermore, an increased understanding of the relationship between TOS and CSM will enable the clinician to inform patients about potential treatment outcomes.

## Author contributions

**Conceptualization:** Xue-Jun Cui and Qi Shi.
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