

# A Patient with Arterial Thoracic Outlet Syndrome Relieved by a Calcium Channel Blocker

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Thoracic outlet syndrome (TOS) results from the compression of the neurovascular bundle, causing neuropathic symptoms and/or ischemic symptoms.<sup>1</sup> Arterial TOS constitutes only 1% of all TOS cases, while it may also be underestimated in the absence of ischemic events or in case they are masked by the neurogenic symptoms.<sup>2</sup>

A 43-year-old woman admitted to the neurology outpatient clinics complaining of a bizarre sensation in her right arm for about 6 months. She had difficulty describing the feeling, but she reported that it was something like a mixture of aching and tingling. She used to awaken from sleep at night because of this bizarre painful feeling. She also noticed that there were undulating attacks of weakness in her right arm, especially becoming evident after using her arm for heavy work. She denied any other associated symptoms in her arm or in any other parts of her body. Her past medical history and family history were unremarkable. She was not on any drugs or substances.

Her systemic examination was normal. In her neurologic examination, the muscle strength, deep tendon reflexes, and sensory examination were all normal.

Further investigations were made with a suspicion of radiculopathy or a spinal pathology. Contrast-enhanced cervical magnetic resonance imaging (MRI) showed diffuse bulging in cervical C5-6 intervertebral disc with minimal fissuring and central protrusion in cervical C6-7 intervertebral disc with a decrease in anterior subarachnoid space (Figure 1). However, the neural foramina were bilaterally free, and there was no pressure on the spinal cord and/or neural structures. Moreover, electromyography did not reveal any abnormal potentials. Cervical radiculopathy and/or spinal pathology was therefore ruled out as a potential contributing factor. Elongation in the spinous process of the seventh cervical vertebral bone was noticed. Contrast-enhanced cranial MRI was also performed to rule out an intracranial pathology, which was normal. She was given analgesic medications and physical therapy with the use of a collar for 1 month, which didn't show any benefit.

She was consulted with a rheumatologist to exclude an underlying vasculitis. A detailed biochemical work-up, including vasculitis markers, protein electrophoresis, rheumatologic and infectious biomarkers, resulted to be normal. Contrast-enhanced thoracic computed tomography (CT) and CT-angiography (Figure 2 and 3) showed normal parenchymal structures of the lung and normal brachiocephalic and subclavian arteries.

She was then consulted with a physical therapist. At her examination, it was noticed that her symptoms were triggered upon elevating her arm above her head. She was asked to keep her right arm superiorly and posteriorly, which she could keep for less than one minute due to the rapid emergent of the ischemic pain and claudication, defined as a painful-like feeling by the patient. She was able to keep her left arm up for much longer without any symptomatology. It was then noticed that her pulse went to 0/4 at this position; in addition, her right palm became pale and cold compared to her left palm. She was clinically diagnosed as having arterial TOS. Putting stress on the neurovascular bundle or Roo's test (elevated arm stress test)<sup>1</sup> to trigger a possible co-existing neurogenic component, and the provocative Elvey test<sup>3</sup> were all negative. She was asked for dynamic Doppler imaging for bilateral upper extremities. It was observed that the third part of the right-sided axillary artery decreased in diameter with the triggering maneuver at first but then ceased completely with a reversed flow showing the same colored signal as the axillary vein. It was learned that she played violin and windsurfed for about 10 years. She also stated that she started anti-aging face and necklace yoga for the last year. Chest x-ray was performed and did not show any extra rib or abnormalities in bony structures around the neck and shoulder (Figure 4). A written informed consent was obtained from the patient who agreed to take part in the study.

The diagnosis of arterial TOS is mainly based on clinical history, physical examination, and different imaging modalities, such as plain thoracic inlet and cervical spine radiography, duplex scan, CT angiography, and conventional angiography. Plain thoracic inlet and cervical spine x-ray graphics may demonstrate cervical rib, anomalous first rib or prominent seventh cervical transverse process. Duplex scan at rest and provocation may demonstrate post-stenotic dilatation, subclavian artery aneurysms, or arterial thrombosis. The CT-angiography at rest and provocation may also contribute to the detection of vascular complications.<sup>4</sup> As an elongated spinous process of the seventh cervical vertebral bone was demonstrated in the patient, and duplex scan at provocation showed reversed flow in the axillary artery, the diagnosis of arterial TOS was made in this patient.

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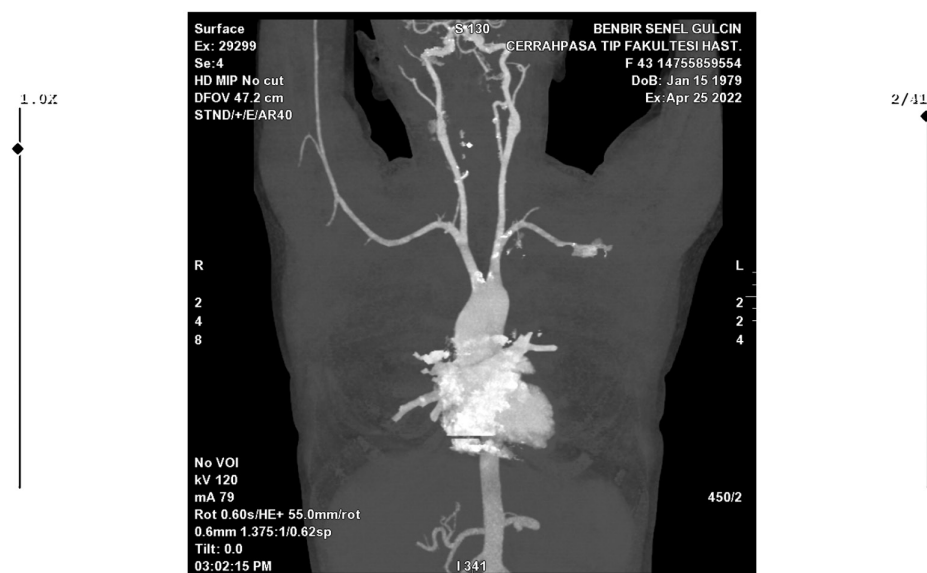


**Figure 1.** Contrast-enhanced cervical magnetic resonance imaging (MRI) of the patient showed diffuse bulging in cervical C5-6 intervertebral disc with minimal fissuring and central protrusion in cervical C6-7 intervertebral disc with a decrease in anterior subarachnoid space. Neural foramina were bilaterally free without any pressure to neural structures.

Upon the diagnosis of arterial TOS, she was consulted with a cardiovascular surgeon and suggested to have surgery for the extraction of the first rib, scalene media, and pectoralis minor muscles. She declined to undergo a major surgery. Then, although a neurogenic component was not detected, empiric lidocaine injection (3.5 mg) was given to the right-sided scalene media muscle, which resulted in a dramatic relief of her symptoms almost within 1 hour. She stated that her symptoms were gone totally for 1 week, and the benefit continued up to 1 month. At the end of 1 month, lidocaine injection was repeated with the same substantial benefit. This benefit was first related to the presence of an undetected neurogenic TOS component, and botulinum injections were planned. One month later, botulinum injections to pectoralis minor, major, and scalene media muscles (a total of 100 IU) were made, but surprisingly it showed no effect at all. At the end of 3 months, with no resolution of the symptoms, another lidocaine injection was given to the right-sided scalene media muscle, which resulted in a similar dramatic relief of her symptoms.

It was discussed that the beneficial effect of lidocaine may be via its sympathomimetic effects causing a local arterial vasodilatation, rather than muscular blocking effects. With this idea, a vasodilator agent potent in arteries, a calcium blocker agent amlodipine (5 mg/day) was given to the patient, which resulted in pronounced alleviation of her symptoms. For 6 months, she is almost symptom-free under amlodipine therapy, and being protective for the triggering maneuvers as much as possible.

There are many clinical studies concerning the efficacy of surgical treatment of TOS, while there are few regarding conservative treatment, and they all include neurogenic TOS or mixed neurovascular TOS. Among nonsurgical interventions, physical therapy including myofascial release maneuvers, myofascial thoracic inlet diaphragm release, muscle strengthening and stretching, lymphatic massage, breathing education, nutrition, and hydration assessment have been shown to be beneficial in neurogenic TOS by reducing symptomatic muscle hypertonicity and myofascial constriction, and alleviating pressure on neurovascular structures.<sup>5,6</sup> Although



**Figure 2.** Thoracic computed tomography (CT) of the patient showed normal parenchymal structures of the lung.



**Figure 3.** 3D representation of thoracic CT-angiography of the patient showed normal brachiocephalic and subclavian arteries.

conservative treatment seems to be effective at reducing symptoms and improving function, outcomes of conservative therapy are hindered by small sample sizes, disparities in inclusion criteria, and different methodologies.<sup>5,7</sup> On the contrary, it has not yet been established whether or not conservative treatment is effective in arterial TOS.

The gold standard treatment for arterial TOS is the surgical decompression or the removal of the offending cause,<sup>8</sup> while some lesser invasive methods are being performed.<sup>9</sup> The axillary approach is mainly suggested for the surgical decompression of neurogenic TOS, the paraclavicular approach is used for the venous TOS, but the supraclavicular approach is almost always used for the arterial TOS, providing an excellent access to all neurovascular structures.<sup>10</sup> While surgical decompression constitutes the major choice of treatment in vascular and/or neurogenic TOS, botulinum toxin injections may also offer short-lasting symptom relief, but only in neurogenic TOS, based on limited quality evidence.<sup>11</sup> Its role in relieving vascular TOS is, however, currently unexploited. Calcium channel blockers, on the other side, result in relaxation in arteries and a decrease in intraarterial pressure, which may explain the alleviation of the patient's symptoms. This case report arises a question for the non-surgical treatment option for the arterial TOS by calcium channel blockers, for the first time in the literature, which deserves to be answered by double-blind, placebo-controlled studies.

**Data Availability Statement:** The data that support the findings of this study are available on request from the corresponding author.

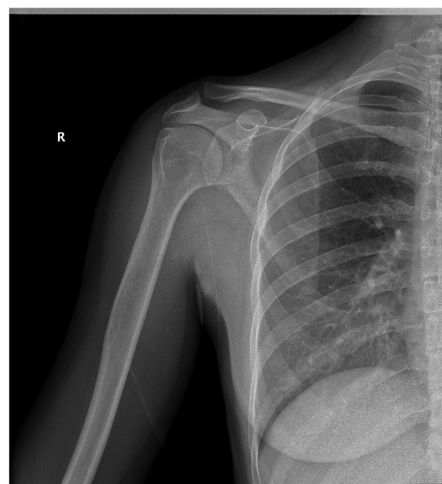
**Informed Consent:** Written informed consent was obtained from the patient who agreed to take part in the study.

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**Figure 4.** No extra rib or abnormalities in bony structures were observed in the chest x-ray of the patient.



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