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Case Report: Thoracic Outlet Syndrome in an excessively trained young olympic athlete

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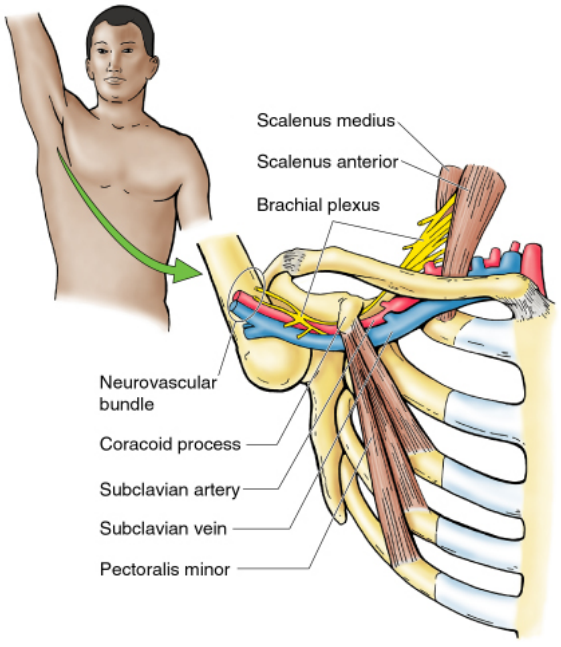
Abstract:
Background: Thoracic outlet syndrome is a set of conditions, first characterized in 1959 by Rogers [1] caused by compression of the neurovascular bundle of brachial plexus and/or subclavian vessels. Among the population it can range in site, type of compressed structures and cause of compression bringing different effects to ones quality of living. One of the possible mechanisms may occur while performing physical exercises including repetitive motions, where overused muscles are causing hypertrophy, which is contributing to the compression.
Case Description: During winter season a 19-year old polish youth national team member of cross-country skiing presented with left subclavian venous thrombosis associated with pain and tingling sensations. Few months after undergoing thrombectomy he presented with burning sensations and decreased strength of left upper limb. Being diagnosed in Switzerland with Thoracic outlet syndrome (TOS) and being informed about the possible outcome of open thorax surgery and associated long rehabilitation he resigned from his olympic career.
Literature review: There are numerous causes of TOS described in the literature, that are subcategorized into neurogenic (nTOS), venous (vTOS) and arterial (aTOS) depending on the main structure responsible for the symptoms. Prevalence of this syndrome also vary, depending on the population, sex, age and frequency of physical exercises performed by individual. Even though nTOS is by far the most common, representing about 95% of cases [2], the overused hypertrophic muscles can relatively often develop venous TOS which is also possible following repetitive motion. Paget–Schroetter disease, also called ‘‘effort thrombosis,’’ involves axillary or subclavian venous thrombosis following strenuous repeated activity with the arms [3].
Purpose and Clinical Relevance: Thoracic outlet syndrome is a condition which can be asymptomatic for a long time, but can also reduce the quality of living, or even lead to systemic disorders like hemorrhages and subsequent fibrosis, that might even become life threatening. Therefore it is essential to suspect it in the differential diagnosis of any possible related case, where other pathologic conditions can be ruled out.

Introduction:
To understand the possible mechanisms leading to the development of thoracic outlet syndrome it is important to distinguish the anatomy of the outlet areas presented in fig.1. There are three compartments where vessels and/or nerves may be entrapped causing the symptoms of TOS. First of them is costoclavicular space which is bordered by subclavius muscle anteriorly and clavicle superiorly. The first rib and anterior scalene muscle form the inferior and posterior borders. Second and also the most lateral compartment is the subcoracoid space with the coracoid process forming the superior boundary, while ribs form the posterior border. Third compartment is the interscalene triangle surrounded by scalene muscles and lying above the first rib. Both subclavian vessels and nerves of brachial plexus passes through all the mentioned compartments, except for the interscalene triangle where subclavian vein goes anteriorly to the muscle.



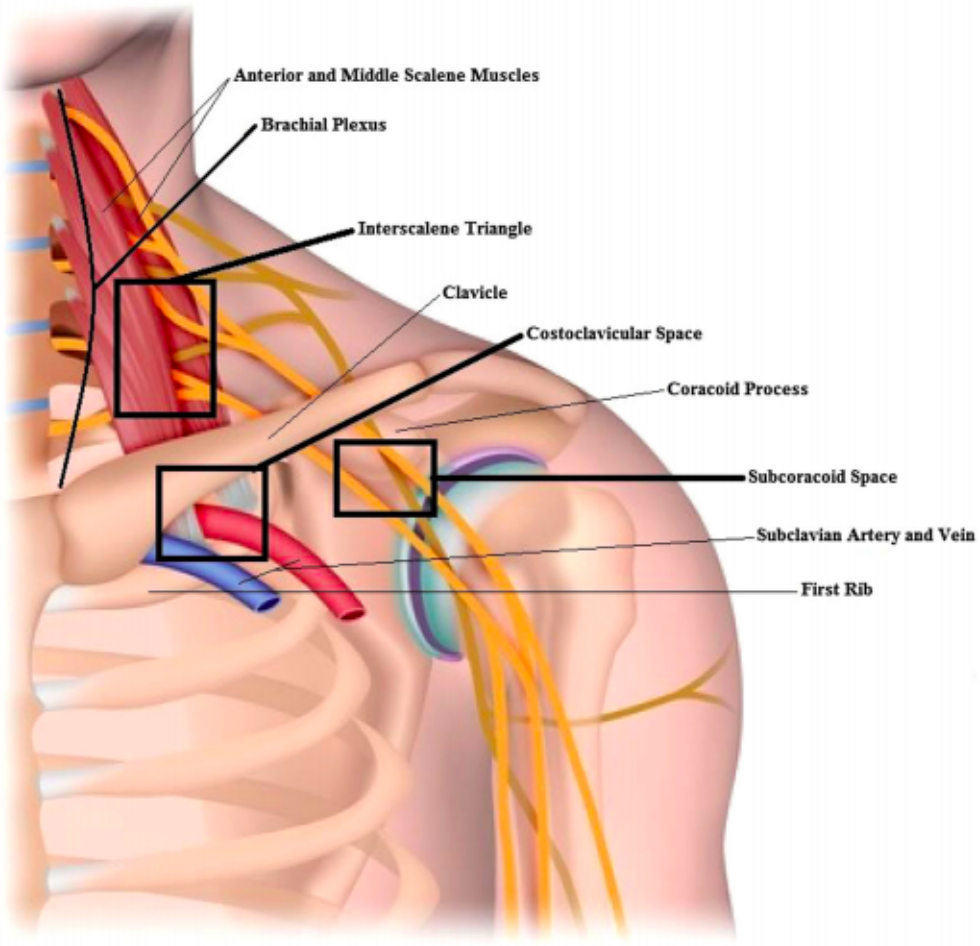


Fig. 1 Thoracic outlet and relevant anatomy

Likewise the anatomic locations of possible entrapment can be different, the etiology also can vary from traumas and injuries, uncontrolled muscle growth caused by specific repetitive movements during physical exercises or anatomic variations. One of such variations is a presence of cervical rib, with prevalence of 1-2% among the population. In most of the cases it remains asymptomatic. However patients with a cervical rib are at higher risk of nTOS, with up to 20% of nTOS cases attributable solely to the presence of a cervical rib [4].
It is also important to exclude other possible causes of compression, which may be caused by malignancy, more precisely by Pancoast tumor. Pancoast tumors, also known as superior pulmonary sulcus tumors, can invade and compress the brachial plexus [5]. Malignancies though, are rare among young people, but shall definitely be excluded in the differential diagnosis.

Case Report:
This case describes a 19 year old male youth national polish member of cross-country skiing. In the beginning of winter season in 2017 year he presented with a varicose vein on his left biceps. The varicose vein was enlarged after physical exercises and accompanied by high fevers. Therefore it disallowed him to continue trainings. In may the same year he was examined in Zakopane. On the angiography it appeared that he is having his left subclavian vein occluded and so it was surgically unblocked. The operation went correctly eliminating the painful varicose vein. However after a few months another problem appeared. Although there wasn't an occlusion any more, during excessive trainings, he started to feel a decrease in his left arm strength, compared to his right arm. On physical examination with his left arm positioned neutrally, his pulse was present and equally strong compared to the contralateral side. However when provocation tests (more later in the text) were applied to his affected side, his pulse drastically decreased and his arm strength weakened. Additionally he experienced a tingling sensation and a hypesthesia. This condition disallowed him to continue trainings once more. Being suspected

with thoracic outlet syndrome he was fully examined. He did an ultrasound scan and later CT of his arm, and later an MRI of his cervical spine. There was no disc disease, nor any additional fibrous bands. Any malignancies were excluded. Taking into account all the mentioned examinations, he was informed about his condition, and proposed an operation of his first rib removal. It turned out that during physical exercises his clavicle and first rib compresses his left subclavian vein in a scissor-like pattern. Additionally, because of his musculature development, the outlet of the subclavian vein was markedly decreased.

Unfortunately during the operation in Switzerland there was a complication which disallowed the total rib excision. His first rib was only partially removed, which decreased pressure put by his bones on the left subclavian vein, but did not fully eliminate it. Post operative rehabilitation lasted two months, during which, patient was not able to perform any routine sport exercises related with cross-country skiing. He was informed about the necessity of a re-operation, if he decided to come back to his full potential of olympic career. However taking into consideration additional operation with possible side effects and another long rehabilitation afterwards, he decided not to undertake it, simultaneously refusing to continue his olympic path career.

Discussion:

Among the possible mechanisms leading to the development of thoracic outlet syndrome, repetitive vertical upper limb movements are important factor possibly leading to both overuse and muscle hypertrophy. It may cause the decrease in the outlet space for neuromuscular bundle of brachial plexus and subclavian vessels. Cross-country skiing is a perfect example of sport with such repetitive movements. Patient described in a case report developed venous TOS which was accompanied by thrombosis. Because of the low susceptibility of this rare syndrome, evaluation of his case was delayed. If he would be examined earlier while the syndrome was developing, it might have been able to combine appropriate treatment faster, and the surgery outcome might also have been better. Unfortunately patient did quit his olympic career after the unsuccessful surgical performance.

Since the first use of term „Thoracic outlet syndrome" there has been a significant development in diagnosis and treatment. It is a rare syndrome reported in several articles with the prevalence among the population of 3-80/1000 [6]. Therefore applying the proper initial treatment is very important for ones future outcomes, and it should be suspected in differential diagnosis of patients with tingling sensations of upper limb, as well as weakening of strength related with physical activities. Diagnosis of the syndrome should be started with provocative tests presented in table 2, which has different sensitivity and specificity, but performed together raise the chance of TOS evaluation.

Venous and arterial TOS are diagnosed by combining clinical presentation, positive results of provocative tests and imaging. For suspected vascular thoracic outlet syndrome, ultrasound imaging maintains high specificity and sensitivity and should be the initial choice, being non invasive and inexpensive. More expensive examinations like CT or MR angiography should be performed prior the surgical operation to provide additional anatomic details, required in the operation theatre. However there is still no established timing of suggested imaging pre- and postoperatively. For example, venograms at 2 weeks after first rib resection and scalenectomy may encounter residual post- surgical inflammation; the precise timing post- surgical venography requires further investigation [7].

After diagnosis of TOS, treatment management should be started, which also vary on timing and sequence. Multimodal treatment should primarily include patient education, physical exercises with TOS- focused muscle strengthening and optional pharmacological intervention of muscle relaxants, NSAIDs and antidepressants.

Physical therapy and conservative management of TOS should persist for at least 4–6 months prior to

Table 2 Common provocative diagnostic tests for thoracic outlet syndrome

Test	Maneuver	Result
ADSON TEST	Affected arm is abducted 30° at the shoulder while maximally extended. While extending the neck and turning head towards ipsilateral shoulder, patient inhales deeply	Decrease or absence of ipsilateral radial pulse
Elevated Arm Stress Test (EAST) or ROOS	Arms are placed in the surrender position with shoulders abducted to 90° and in external rotation, with elbows flexed to 90°. Patient slowly opens and closes hand for 3 min	Precipitates pain, paresthesias, heaviness or weakness
Upper Limb Tension Test (ULTT) or ELVEY	Position 1: arms abducted to 90° with elbows flexed Position 2: active dorsiflexion of both wrists Position 3: head is tilted ear to shoulder, in both directions	Positions 1 and 2 elicit symptoms on the ipsilateral side, while position 3 years elicits symptoms on the contralateral side

consideration of surgical intervention. However, for patients with arterial or venous TOS, the initial intervention is most often surgical [8]. Even though surgical outcomes of decompression are very positive, reported as „excellent" results in 95% [9], preoperative management is essential to lower the chance of complications, such as life threatening pneumothorax. Nowadays new approaches to surgical advancements are being developed. One of such is a robotic-assisted and endoscopic- assisted trans-axillary technique with potential benefit, the latter aiming to decrease risk of pneumothorax [10].

Thoracic outlet syndrome is only one of many rare conditions which may not only decrease the quality of ones living, but also often disable sport professionals, leading to premature end of career; like the one presented in this case report. Therefore improvement of diagnosis and development of well established treatment regimens, is essential for the future of sport medicine.

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