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Differentially diagnosing chronic upper limb paresthesia in a 24-year-old patient: is thoracic outlet syndrome the culprit? A case report

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Objective: *To describe the differential diagnosis, diagnosis, and chiropractic management of a case of chronic upper extremity paresthesia.*

Clinical features: *A 24-year-old woman presented with recent neck stiffness, along with a primary complaint of chronic upper extremity paresthesia and hand weakness of insidious onset.*

Intervention and outcome: *Results of previous electro-diagnostic and advanced imaging studies were combined with clinical assessment to diagnose thoracic outlet syndrome (TOS). Discontinuing after five weeks of chiropractic management, the patient reported significant improvement of paresthesia but less improvement of her hand weakness.*

Diagnostic différentiel de la paresthésie chronique des membres supérieurs chez une patiente de 24 ans: le syndrome du défilé thoraco-brachial est-il en cause? Un rapport de cas.

Objectif : *Décrire le diagnostic différentiel, le diagnostic et la prise en charge chiropratique d'un cas de paresthésie chronique des membres supérieurs.*

Caractéristiques cliniques : *Une femme de 24 ans s'est présentée avec une raideur de la nuque récente, ainsi qu'avec une plainte primaire de paresthésie chronique des membres supérieurs et de faiblesse de la main d'apparition insidieuse.*

Intervention et résultats : *Les résultats d'un électrodiagnostic antérieur et d'examens d'imagerie avancée ont été combinés à une évaluation clinique pour diagnostiquer un syndrome du défilé thoraco-brachial. Après cinq semaines de traitement chiropratique, la patiente a signalé une amélioration significative de ses paresthésies, mais une amélioration moindre de la faiblesse de sa main.*

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Summary: *Several etiologies can give rise to symptoms in common with TOS. It is imperative to rule out mimicking conditions. A battery of clinical orthopedic tests has been proposed in the literature for the diagnosis of TOS but with reported questionable validity. As a result, TOS is mostly a diagnosis of exclusion. Chiropractic treatment shows potential for effective management of TOS, but research is required.*

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KEY WORDS: chiropractic, thoracic outlet syndrome

Résumé : *Plusieurs étiologies peuvent donner lieu à des symptômes communs avec le syndrome du défilé thoraco-brachial. Il est impératif d'exclure les affections mimétiques. Une batterie de tests orthopédiques cliniques a été proposée dans la littérature pour le diagnostic du syndrome du défilé thoraco-brachial, mais leur validité est discutable. Par conséquent, le syndrome du défilé thoraco-brachial est le plus souvent un diagnostic d'exclusion. La chiropratique est susceptible de permettre une prise en charge efficace du syndrome du défilé thoraco-brachial, mais des recherches s'imposent.*

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MOTS CLÉS : chiropratique, syndrome du défilé thoraco-brachial

Introduction

Over 30 years ago, thoracic outlet syndrome (TOS) was a controversial medical diagnosis.[1] Today, it seems to be more understood and surprisingly more prevalent than once thought.[2] Over the years, it had been termed costoclavicular syndrome, cervical rib syndrome, scalenus anticus syndrome, subclavius tendon syndrome, or musculus pectoralis major syndrome by various authors.[3,4]

This previous controversy might be related to the complexity of TOS involving several different anatomical structures (brachial plexus, subclavian artery, and subclavian vein) that are compromised in different anatomical spaces (scalene triangle, pectoralis minor and costo-clavicular). Thus, the clinical picture of TOS remains challenging to diagnose because it has been reported to cause an array of signs and symptoms.[5] Perhaps it is also because of the lack of valid clinical tests[6], relegating it to a diagnosis of exclusion. Therefore, it is important to rule out several potentially serious conditions that can compromise the neurovascular bundle of the upper extremities.

The purpose of this case report is to review a clinical presentation of chronic neuropathic symptoms affecting the upper extremity of a 24-year-old woman. Electrodiagnostic investigation and advanced imaging suggested this case was compatible with the neurogenic type of TOS. Several differential diagnoses along with their clinical examination are presented to help the astute clinician rule out important conditions before making a clinical diagnosis of TOS.

Case report

A 24-year-old female, right-handed building inspector presented with a main complaint of muscular atrophy in her right hand and chronic paresthesia of her right little finger. She also reported stiffness in the neck and right trapezial region. The paresthesia in her right little finger began insidiously over the past two years. She described her paresthesia as a constant tingling sensation affecting her entire fifth digit and more recently the medial aspect of her forearm to the elbow. Sleeping on her right side with her right arm under her pillow aggravated the tingling sensation in the fifth digit.

For the past two months, she noticed some muscular wasting involving the palmar aspect of her right thumb (Figure 1), as well as weakness with grip strength. As a result, she had difficulty grasping objects with her right hand, such as a glass of water. Also, for the past couple of months, she had noticed some neck stiffness, more so on the right than the left. She had no recent history of trauma to her neck or right upper extremity. However, she was involved in a highway rollover motor vehicle accident (MVA) three years prior without any apparent injur-

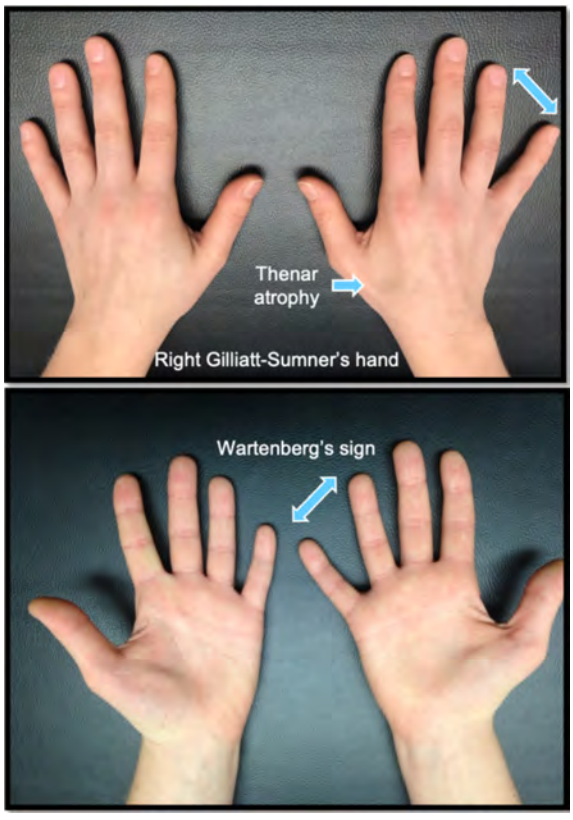

Figure 1.
Gilliatt-Sumner's hand and Wartenberg's sign

ies or hospitalization. She was in good general health, a non-smoker, and did not take any medication.

Two months prior to her presentation, she consulted with a neurologist regarding the tingling in her right little finger. The neurologist's report indicated significant muscular atrophy and weakness (3/5) of the right first interosseus muscle, abductor pollicis brevis and abductor digiti minimi. Deep tendon reflexes of the upper extremities were symmetrically 2+, except for a diminished right triceps reflex (1+). Hypoesthesia was noted in the right C8-T1 distribution, especially in the ulnar nerve territory. Tinel's sign was absent at the wrist (over the flexor retinaculum and Guyon's canal) and elbow (cubital canal). At that time, she underwent electromyography of the right upper extremity which revealed abnormal electrical activities of the median and ulnar nerves, with significant denervation of the abductor pollicis brevis and first interosseus muscle of the right hand.

To rule out a cervical radiculopathy, MRI of the cervical spine and brachial plexus without contrast was ordered by the neurologist. The MRI showed no cervical disc herniation, lateral/foraminal stenoses or cervical ribs. Though the diagnosis remained uncertain, no further investigations were planned. She decided to seek chiropractic care to alleviate her symptoms.

On presentation at the chiropractic clinic, the Neck Disability Index (NDI) was rated at 18%, indicating only a mild neck pain-related disability.[7] She reported the intensity of her tingling ranging from 6 to 8/10 on a visual analogue scale (VAS).[8] The Disabilities of the Arm, Shoulder, and Hand Score (QuickDASH) was 48%, suggesting a moderate amount of disability with using her right hand. The QuickDASH questionnaire measures an individual's ability to complete tasks and absorb forces, and the severity of symptoms for any patient with upper extremity musculoskeletal disorders.[9]

Physical examination

Examination at the Outpatient Student Chiropractic Clinic revealed hollowing of the intermetacarpal spaces and the web space of the right hand giving the appearance of the Gilliatt-Sumner's hand (Figure 1). The muscular atrophy was especially noticeable at the thenar eminence and, to a lesser extent, the hypothenar eminence. Wartenberg's sign was present with the right fifth digit maintained in abduction position at rest suggesting weakness of the adducting palmar interosseous muscle innervated by the ulnar nerve (Figure 1).[10]

She was able to make a fist and there was no claw hand deformity at rest or Benediction hand with a fist. There were no colour or temperature changes in the right upper extremity. Allen's test to assess blood flow in the right upper extremity was normal.

Neurological examination revealed hypoesthesia of the right fifth finger with two-point discrimination at 10mm (normal = < 6 mm), as well as diminished light touch sensation along the medial border of the forearm to the elbow. Two-point discrimination has been shown to be a valid test in routine examination of suspected nerve injuries of the hands and fingers.[11] Strength testing revealed 4/5 strength of the flexor digiti minimi, abductor digiti minimi and abductor pollicis brevis. Froment's sign was positive for weakness of the adductor pollicis, innervated by the ulnar nerve.[10] Deep tendon reflexes of the upper ex-

tremities were symmetrically 2+. Plantar responses were down going and ankle clonus was absent.

Cervical spine active range of motion was full in all directions. Forward neck flexion revealed some discomfort at the back of the neck. Side neck flexion caused contralateral neck pain. Cervical extension was pain-free. Spurling's test was negative for reproducing or aggravating the right upper extremity symptoms. Sustained maximal elbow flexion (60 seconds) increased the tingling in the right fifth digit. However, tapping over the right Guyon's canal, cubital tunnel or supraclavicular fossa did not reproduce her tingling sensation along the medial forearm and fifth digit. Upper limb neurodynamic test (ULNT-Ulnar)[12], which is considered to stress the ulnar nerve, aggravated the tingling in the right little finger and caused a burning sensation in the right lower neck area. The EAST (Elevated Arm Stress Test or Roos test) provocation manoeuvre for TOS[13] was performed for three minutes without exacerbation of symptoms. Hyperabduction and Wright's tests, which are included in the Gillard's provocation tests for TOS[13], did not reveal any diminution of the right radial pulse or reproduction of tingling in the right fifth digit. Muscle tenderness and hypertonicity were noted at the right scalene and pectoralis minor muscles. Spinal joint palpation revealed restriction of C7 on left rotation and restricted movement of the first rib on the right side. Range of motion of the right shoulder was full and pain free in all planes.

Diagnosis

In view of neurological signs and symptoms of the right hand on clinical examination, abnormal electrical activities of the median and ulnar nerves from a recent EMG study, and a negative cervical spine MRI, a diagnosis of chronic neurogenic TOS was given to her condition.

Intervention and outcome

The persistent tingling over the right medial forearm nearly resolved during the course of five weeks of care (twice per week). She received a total of ten chiropractic treatments consisting of spinal manipulation therapy using diversified adjusting technique to C7 vertebra and first rib articulations to restore motion. Hypertonic scalene and pectoralis minor muscles were provided soft tissue therapy (trigger point therapy, hold-relax with antagonist contraction (PNF), and deep tissue massage), as well as a daily home program of stretching exercises. Also, she was given a home program of isometric exercises to strengthen the intrinsic muscles of her right hand. She reported an overall improvement of 50% for the significant improvement of her tingling sensation but none regarding the return of strength of her right hand. The tingling sensation of the right little finger had mostly dissipated, although some hypoesthesia at the tip of the fifth digit as well as the muscular atrophy of both the thenar and hypothenar eminences persisted. The complaints of neck stiffness and trapezial pain had resolved completely. Unfortunately, the NDI and QuickDASH questionnaires were not reassessed at self-discharge or upon the telephone interview at two years follow-up.

In the middle of winter, the patient had to travel 45 minutes each way to attend her chiropractic treatments and, with consideration of the cost of chiropractic treatments, eventually decided to discontinue her care. She was referred to her family physician for a referral for a physiotherapy and strengthening program, which would have no cost to the patient. At follow-up two years later, she reported no change in her condition. She did not go for physiotherapy after all and occasionally receives chiropractic treatments closer to her home. She has continued to work full-time as a building inspector.

Discussion

Based on the clinical presentation alone, the diagnosis of neurogenic TOS is very challenging. A systematic review by Sanders *et al*.[5] revealed a wide diversity of symptoms in neurogenic TOS: upper extremity paresthesia (98%), neck pain (88%), trapezius pain (92%), shoulder and/or arm pain (88%), supraclavicular pain (76%), chest pain (72%), occipital headache (76%), and paresthesia in all five fingers (58%), the fourth and fifth fingers only (26%), or the first, second, and third fingers (14%). Any of these symptoms can be caused by various musculoskeletal dysfunctions of the cervicothoracic region and upper extremity, instead of or concurrently with TOS. Depending on the predominant structure and site of compression involved (Figure 2), TOS can be divided into three subtypes, namely neurogenic TOS (nTOS, compression of brachial plexus), arterial TOS (aTOS, compression of subclavian artery) and venous TOS (vTOS, compression of subclavian vein).[2] Mechanisms of compression have been reported to include fibrous muscular bands, cervical

ribs, and pressure in the inter-scalene triangle space, the pectoralis minor space and the costo-clavicular space.[14]

Arterial TOS is characterized with coolness and pallor in the hand (similar to Raynaud's phenomenon), upper limb ischemia and digital ulceration, while vTOS presents with upper limb swelling and cyanotic discoloration.[15] The venous and arterial subtypes of TOS affect only 3% and <1% of all TOS patients, respectively.[16] The diagnosis of arterial and venous TOS can be confirmed by angiography.[17]

The most common form by far is believed to be the neurogenic type (nTOS), which may account for >90% of all TOS cases.[2] The brachial plexus trunks or cords, originating from nerve roots C5 to T1, are affected in nTOS and can present variable patterns of upper limb weakness, numbness and paresthesia.[15] The paresthesia can involve all five fingers but commonly is worse in the ulnar nerve distribution involving the fourth and fifth fingers.[18]

Neurogenic TOS can be further divided into true or disputed nTOS, with the latter representing 95 to 99% of all neurogenic cases.[19] The symptoms of true and disputed nTOS are largely the same, though objective findings from motor nerve conduction studies and needle electromyography are notably negative in disputed nTOS.[20] Kim *et al*.[21] recently confirmed that true nTOS is predominantly a lower roots/trunk brachial plexopathy involving T1 with clinical and electrodiagnostic features that are largely the same.

Clinical features of the reported case

The case presentation has many characteristics of a true nTOS involving the medial cord of the brachial plexus derived from nerve roots C8 and T1:

1) She is a 24-year-old female with unilateral chronic paresthesia of her right fifth digit. This seems to fit the targeted population for nTOS because teenaged to 60-year-old females are most often affected by true nTOS.[2]
2) The presence of the Wartenberg's sign of the right fifth digit, which is an involuntary abduction of the fifth finger due to a lack of opposition action from the extensor digiti minimi, suggests chronic ulnar nerve palsy. A positive Froment's sign and a positive maximal elbow flexion test further warranted the likelihood of an ulnar nerve lesion at the level of the elbow. However, adding to the complexity of the clinical examination findings, other tests like Tinel's sign at the cubital canal or even at the wrist was absent. Also, there was no acquired claw hand deformity to suggest a simple ulnar nerve palsy. Consequently, the wide range of neurological deficit had to involve different motor fibres, at the level of the brachial plexus.
3) Involvement of the medial cord (C8-T1) was demonstrated from the electromyography study which revealed objective findings of abnormal electrical activities of the median and ulnar nerves, with significant denervation of the abductor pollicis brevis and first interosseus muscle of the right hand that could not be explained by MRI of the cervical spine and brachial plexus.
4) The EMG findings were in keeping with the diffuse muscular atrophy of the right hand giving the appearance of the Gilliatt-Sumner's hand. The Gilliatt-Sumner's hand is a clinical presentation that includes atrophy of the abductor pollicis brevis (median nerve), interosseous (ulnar nerve) and abductor digiti minimi (ulnar nerve) muscles, making it a strong diagnostic indicator of nTOS.[22]
5) Three different types of scale ratings were used.
   a. She scored 18% on the NDI, suggesting only a mild neck pain-related disability. Therefore, it was less likely that her symptoms were directly from a specific neck condition. She had a rollover motor vehicle accident three years prior to the onset of her arm symptoms, but she denied having any sequelae from the MVA.
   b. A visual analogue scale was used to rate the intensity of the paresthesia affecting her entire right fifth digit and the medial aspect of her forearm, which was rated as relatively high at 6 to 8 out of 10. This confirmed the presence of her paresthesia in a distribution consistent with the C8 dermatome, a characteristic of nTOS.
   c. Her ability to use her right hand was 48% on the QuickDASH, suggesting a moderate amount of disability. This could be attributed to the diffuse muscular atrophy of the right hand (Gilliatt-Sumner's hand). The literature indicates that the Quick DASH can be used to measure disability/symptom severity in a variety of upper extremity disorders.[9]

As shown in this case report, several conditions must be excluded prior to establishing a diagnosis of nTOS. The

clinical findings that can mimic nTOS are also consistent with conditions such as cervical radiculopathy, Pancoast tumor, cubital canal syndrome, arcade of Struthers syndrome and Guyon canal syndrome. Consequently, a thorough history, clinical examination, and investigation to exclude similar presenting conditions can increase the accuracy in making the diagnosis of nTOS.[20] These conditions are summarized below with their respective clinical examination findings (Table 1).

Table 1.
Conditions that can mimic neurogenic Thoracic Outlet Syndrome

Condition	Signs and symptoms	Clinical examination	Ruled out in this case report
Cervical spine radiculopathy involving C8 and T1 nerve roots	Neck pain, scapular/periscapular pain and diminished cervical ranges of motion (ROM) are common clinical findings.[38] Loss of dermatomal sensation to the medial aspect of the forearm (C8) or arm (T1), sometimes with diminished deep tendon reflexes and unilateral hand muscle weakness, atrophy or fasciculation in a myotomal distribution.[39]	Combination of Spurling's cervical compression, axial cervical traction, and an Arm Squeeze Test[40] to increase the likelihood of a clinical diagnosis of cervical radiculopathy. Combined results of four negative upper limb neurodynamic tests (ULNT-1-2-3-4) and Arm Squeeze Test could rule out cervical radiculopathy.[12]	Cervical ROM unaffected. Loss of dermatomal sensation to the 5th digit and medial aspect of the forearm (C8) but not the arm (T1).[41, 42] Negative Spurling's test. Reflexes of the upper extremities were symmetrical at 2+. MRI of the cervical spine was unremarkable.
Pancoast tumour Tumour growth and local invasion may lead to a combination of signs and symptoms (Pancoast-Tobias syndrome), the latter presenting several similarities with the clinical presentation of nTOS or C8-T1 radiculopathy (e.g., radiating pain to the neck, the medial aspect of the arm and forearm as far as the wrist).[43]	Middle-aged patient presenting with radicular-like symptoms should be examined neurologically for Horner's syndrome; miosis, partial ptosis and hemifacial anhidrosis.[44] Red flag signs and symptoms of lung malignancy (e.g., cough, hemoptysis, dyspnea, fever, weight loss) may also be seen in the later stages of the disease.[41, 45]	The presence of one or more signs characterizing Pancoast-Tobias syndrome (i.e., Horner syndrome, upper arm edema, paresthesia, weakness in the intrinsic hand musculature), combined with tumour localization through imaging, enable this condition to be distinguished from nTOS or degenerative cervical spine disorders.[43]	Patient is young and did not present with Horner's syndrome. There were no constitutional signs and symptoms.
Ulnar neuropathy The ulnar nerve provides sensory innervation to both the palmar and dorsal aspect of the medial half of the fourth finger, the entire fifth finger and to the ulnar border of the hand.[42]	The typical patient presentation for cubital tunnel syndrome in the early phase involves numbness/paresthesia of the 4th and 5th digits[10] with weakness or atrophy of the intrinsic muscles of the hand in more chronic cases[46]. Occasionally, pain is experienced along the course of the ulnar nerve from the posteromedial elbow into the ulnar forearm or hand.[47] At the level of the wrist an ulnar nerve compression in the ulnar tunnel (Guyon's canal) is characterized by loss of sensation with or without motor dysfunctions, affecting the hypothenar region and the ventral aspect of the fourth and fifth digits.[42, 48] In chronic cases, ulnar nerve compression in the cubital tunnel may lead to paralysis of lumbricales and interossei resulting in acquired claw hand deformity (i.e., hyperextension at the metacarpophalangeal joints and flexion at the proximal and distal interphalangeal joints of the fourth and fifth fingers).[49]	Impaired sensation can be quantified with two-point discrimination examination.[11] Tinel's percussion test can be done over Guyon's canal (ulnar tunnel) and over the cubital tunnel. The latter may be positive for creating a radiating sensation along the ulnar border of the forearm into the hand.[10] The sustained elbow flexion test with the elbow held in full flexion with the wrist in extension may also be positive (reproducing paresthesia and pain). [50] In chronic cases with the acquired claw hand deformity, the patient will still be able to make a fist. In cases of severe and chronic compressive neuropathy of the ulnar nerve, hypothenar atrophy may be present, in addition to positive Froment's and Wartenberg's signs.[10]	Patient's presentation and clinical findings were indicative of mixed neuropathies: Electromyography confirmed abnormal electrical activities of the ulnar and median nerves. Distribution of the paresthesia (fifth digit), muscular atrophy of both the thenar and hypothenar eminences (Gilliatt-Sumner's hand), hand weakness, Wartenberg's and Froment's signs were positive for ulnar neuropathy. However, Tinel's tests were negative along the trajectory of the ulnar nerve (cubital tunnel and Guyon's canal). Although sustained maximal elbow flexion test and neurodynamic tests were positive, there was no claw hand deformity. Unfortunately, the scratch collapse test, which has been described as a useful examination manoeuvre for the evaluation of median and ulnar neuropathy (carpal and cubital tunnel syndromes), was not performed. According to Cheng *et al*.[51] the scratch collapse test is more sensitive than the Tinel's sign. As such, it is possible that our case had a mix of ulnar neuropathy and nTOS.

Condition	Signs and symptoms	Clinical examination	Ruled out in this case report
Brachial plexopathy injuries (BPI) Neuropraxia, axonotmesis, and neurotmesis are the three main classifications of nerve injuries, with neurotmesis being the most severe form of nerve injury.[52] The neuropraxia form of nerve injury is most seen in entrapment neuropathies or pressure palsies, usually with complete recovery within days to weeks.[53]	The signs and symptoms of a BPI depend on the injury's location and extent. In American football, traumatic upper trunk brachial plexopathy injuries, also known as a cervical stinger or burner, is the most common upper extremity neurologic injury, most commonly involving the upper trunk.[54]	Lower brachial plexus lesions, affecting the C8-T1 nerve roots, can mimic the clinical presentation of nTOS, resulting in sensory (i.e., pain, numbness or tingling) and motor (i.e., weakness or paralysis) deficits of the forearm flexors and the intrinsic muscles of the hand, without corresponding to a peripheral nerve distribution.[55, 56] As the sympathetic chain arises from the C8 and T1 nerve roots, damage at this point may also result in Horner's syndrome[55, 56], making it important to assess for P.A.M. (Ptosis, Anhidrosis and Miosis).	Contrary to BPI, the patient's symptoms were not related to a history of traumatic onset.
Neuropathic pain "Pain caused by a lesion or disease of the somatosensory nervous system"[57].	Chronic neuropathic pain is more frequently observed in women above 60 years old[58, 59], and is characterized by spontaneous pain and increased pain sensitivity to painful (hyperalgesia) and nonpainful (allodynia) stimuli, often causing major suffering and disability.[57, 58, 60, 61]	Screening tools have been developed and validated to help identify it in clinical practice.[58] Pain medications might be reported by the patient; recent medical practice guidelines for chronic neuropathic pain recommend Duloxetine, Venlafaxine and Gabapentine as first-line treatments.[62, 63]	The patient did not report any pain, allodynia or hyperalgesia in her upper extremity, only paresthesia mainly of her little finger. She did not receive anti-neuropathic medication. Based on the QuickDASH she was not in the category of high disability.
Progressive neurological disorder such as: Amyotrophic Lateral Sclerosis	Clinically Definite ALS is defined on clinical evidence alone by the presence of UMN (pathologic spread of reflexes), as well as LMN signs (weakness, atrophy and fasciculations), in the bulbar region and at least two spinal regions or the presence of UMN and LMN signs in three spinal regions.[64]	ALS starts with limb weakness in about 2/3 of patients, while the remaining 1/3 have bulbar weakness causing dysarthria and dysphagia.[65] Neurological examination reveals weakness, atrophy and fasciculations, as well as hyperreflexia and increased tone in the same motor segment often combined with an extensor response to plantar stimulation.[65]	Muscular atrophy was noticeable in the thenar and hypothenar eminences of the right hand without apparent fasciculations. Hypoesthesia was limited to the 5th digit. Strength testing showed weaknesses (4/5) of the abductors of the thumb and 5th digit. Deep tendon reflexes of the upper extremities did not show any hyperreflexia and were symmetrical at 2+. Furthermore, there were no UMN signs and no dysarthria or dysphagia.

Legend: *ROM - Ranges of motion (ROM), ULNT – Upper Limb Neurodynamic Test, MRI – Magnetic Resonance Imaging, BPI - Brachial plexopathy injuries, ALS – Amyotrophic Lateral Sclerosis, UMN – Upper Motor Neuron, LMN – Lower Motor Neuron*

Diagnosis of Thoracic Outlet syndrome

Orthopedic examination for Thoracic Outlet Syndrome

Depending on the type of TOS suspected, diagnostic investigations can vary, necessitating a good history taking and clinical examination.[15] Thus far, the scientific literature does not offer any valid orthopedic test to clinically confirm the diagnosis of the different types of TOS.[6] Moreover, it is important to rule out a cervical brachial pain syndrome, such as a cervical radiculopathy, cubital tunnel syndrome, carpal tunnel syndrome, and Guyon's canal syndrome with a neurological examination.

On examination, signs of swelling and cyanotic discoloration may be in keeping with Paget-Schroetter syndrome, also called "effort thrombosis," seen in vTOS (Figure 2). In other instances, Raynaud's phenomenon, upper limb ischemia, and digital ulceration may be more in keeping with compression of the subclavian artery (aTOS).[23] However, any swelling of the upper extremities or changes in skin colour, texture and temperature can also suggest complex regional pain syndrome, but

typically the differentiating hallmark of this condition is excruciating pain, even to light touch.[24] Chronic nTOS might show muscular atrophy of the thenar eminence, hypothenar eminence, and the interossei suggestive of the Gilliatt-Sumner's hand.[25]

It has been suggested that provocative manoeuvres can add weight to a suspected diagnosis of TOS, but alone their utility is questionable.[15] Commonly used provocative manoeuvres have been incorporated into the Gillard's clinical prediction rule, which includes a cluster of five provocative manoeuvres (Adson's, Hyperabduction or Eden's, Wright's, EAST, and Tinel's sign over the supraclavicular fossa). To increase the specificity to 84% all five tests must be positive.[13]

The structures which can cause TOS (hypertonic scalene or pectoralis minor muscles, cervical ribs) can be detected by palpation. Joint movement restrictions can be significant in the cervico-thoracic region, as well as the costovertebral joint of the first rib. Thus, the experienced clinician should be able to diagnose TOS by excluding mimicking conditions through the history (including mechanism of onset of the condition, type of symptom and its localization) as well as the observation of subtle clinical signs.

Imaging

Clinicians must often rely on imaging modalities in combination with provocative tests since little evidence supports the validity of a single orthopedic test for the diagnosis of TOS.[6] Since electromyography results are commonly normal in patients with nTOS (i.e., ''disputed nTOS'')[26], imaging is used to rule out other neurologic conditions (e.g., ulnar or median neuropathies, cervical radiculopathy)[27].

The American College of Radiology (ACR) criteria for imaging in the diagnosis of TOS suggest that chest radiography should be considered as a primary imaging modality, given its low cost, ease of access and safety profile.[28] This type of imaging enables the identification of osseous abnormalities such as cervical ribs, first rib defects, other congenital malformations or soft tissue lesions[29], as well as pathologies such as lung tumors, which can trigger symptoms of nTOS. MRI without contrast is also recommended for the diagnosis of nTOS, typically performed during provocative positioning (e.g., patient's arm in abduction). Ultrasonography (US) can be useful

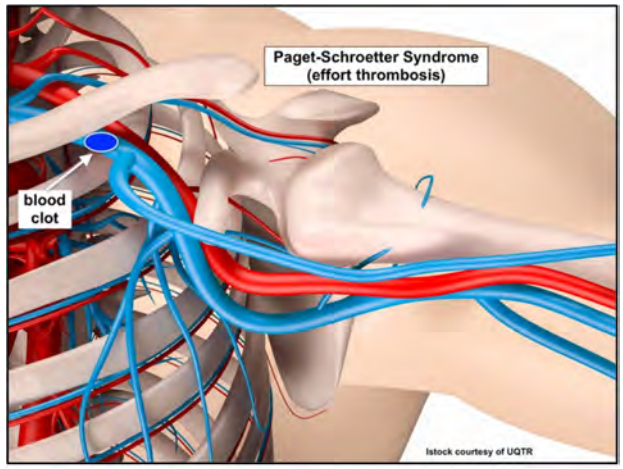

Figure 2.
Paget-Schroetter syndrome

in the identification of fibromuscular bands that may cause compression of the lower brachial plexus[30] or can be used to rule out more distal compression of peripheral nerves by similar anatomical structures (e.g., Arcade of Struthers)[31, 32].

Outcome measures

To date, there are no validated scales or questionnaires that have been developed specifically to measure functioning or quality of life in patients diagnosed with TOS. Recent studies assessing the impact of conservative approaches to treat TOS[33, 34] have used the Disabilities of the Arm, Shoulder, and Hand (DASH) questionnaire or its shortened version (QuickDASH), both validated and designed to monitor upper extremity function over time[9]. A patient with no disability will score 0; one who has maximal disability will score 100. Also, the validated Cervical Brachial Symptom Questionnaire (CBSQ) can be used for widespread sensory and motor findings that would indicate a condition such as cervical radiculopathy, carpal tunnel syndrome or ulnar neuropathy. It is used by adding together the sum of all 12 numerical questions. A patient with no disability will score 0; one with maximal disability will score 120.[35] Lately, a group of experts has worked on the development of a thoracic outlet syndrome index (TOSI), combining several items from validated questionnaires.[36] Although preliminary results of this study are promising, further research is warranted to support its validity and promote its use.

Chiropractic management

A recent published scoping review on exercise rehabilitation for neurogenic TOS did not find any randomized controlled trials.[37] They found several treatment options such as analgesic and anti-inflammatory medication, botox injections, hot/cold therapy, electrophysical modalities (TENS, US), manual therapy, orthoses, night splinting, and patient education regarding ergonomics and postural awareness. They concluded that the clinical rationale proposed by most authors involved postural correction and decompression of the thoracic outlet via restoring proper muscular balance. Rehabilitative exercises for strained musculature would commonly be prescribed. Luu *et al*.[37] reported inconsistency across studies regarding exercise dosage.

Since these treatment approaches target dysfunctional muscles and articulations, and not directly nerves or blood vessels, the question arises: Does differentiation of TOS into neurological or arterial subtypes serve a purpose in the conservative management of TOS? The nerves and blood vessels travel through the thoracic outlet in close proximity (Figure 3). It is unlikely that different degrees of muscle tension would selectively compress, for example, arterial rather than nervous structures. Furthermore, if aTOS compromises the arterial blood flow through the vasa nervorum of the nerves coursing through the thoracic outlet, should the resultant symptomatology be diagnosed as nTOS or aTOS? If a manoeuvre compromising the thoracic outlet and eliciting or exacerbating upper extremity symptoms, whether with or without attenuation of the radial pulse, were considered positive, it could make the clinical assessment and diagnosis of TOS simpler, more valid, and possibly more reliable.

In the case presented, the diagnosis of chronic neurogenic thoracic outlet syndrome (nTOS) was based on the normal MRI that excluded the presence of a cervical disc herniation, Pancoast tumor, fibrous band, cervical rib or other space occupying lesions. While the EMG study indicating abnormal electrical activities in both the median and ulnar nerves was consistent with disturbance at the lower trunk of the brachial plexus, the appearance of a Gilliatt-Sumner's hand suggested C8 and T1 nerve root involvement. Though the natural history of TOS is not well defined, the chronicity of the symptoms, abnormal electromyography, and muscular wasting of the hand

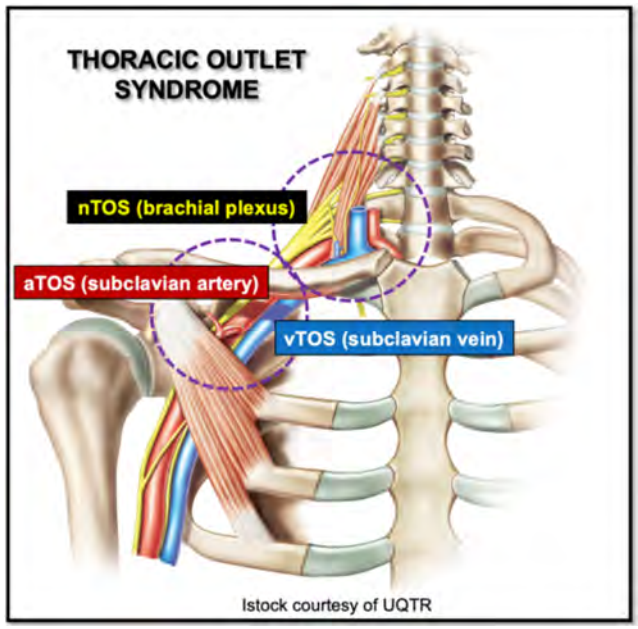

Figure 3.
Relevant anatomy of three types of thoracic outlet syndrome

strongly suggested that the patient was not likely to recover on her own.

Numerous mechanisms associated with TOS have been reported, including motor vehicle accident, repetitive motions and anatomic variations.[20] Our patient was involved in a roll-over motor vehicle accident three years prior to receiving chiropractic treatments. Although she denied any sequelae post-MVA, the role of this trauma in initiating musculoskeletal dysfunctions which caused her subsequent nTOS symptoms cannot be ruled out.

Following a trial of ten sessions of manual therapy (joint mobilization, manipulation, and soft tissue massage) for a diagnosis of neurogenic TOS, the patient reported a 50% improvement, with no tingling along the medial border of the forearm and resolved neck stiffness. Manual therapy seems to have had an effect by relaxing the scalene and pectoralis minor muscles, which could have resulted in lesser compression of the lower cord of the brachial plexus, thus improving her paresthesia. However, over the relatively short period of treatment, the hand muscular atrophy and hypoesthesia at the tip of the fifth digit persisted. Two years later, she reported no further improvement in her condition. For comparison, in a

case series of nine TOS patients surgical decompression of a fibrous cervical band was beneficial for the relief of hand pain and paresthesia and slight recovery of power on the affected hand, but muscle wasting in the hand appeared to be unchanged up to eight years later.[25] Given the opportunity to provide more chiropractic treatments to our patient, it is possible that more improvement could have been obtained with regards to the paresthesia in the fifth digit, but improvement of the muscular atrophy would be unlikely.

Surgical management

The patients who are likely to benefit from a surgical intervention are those with troublesome pain and paresthesia, and those with a progressive history of weakness and muscle wasting.[25] Upon the telephone interview two years post-treatment, our patient indicated that a surgical intervention was not being considered at the present time. Typically, surgical interventions involve the division of a fibromuscular band that may cause compression of the lower brachial plexus or a resection of a rudimentary cervical rib.

Summary

Due to the three subtypes of TOS (aTOS, vTOS and nTOS) as well as true versus disputed TOS, its diagnosis remains a challenge. The difficulty arises not only from the plethora of clinical presentations but also from the poor reliability of most orthopedic provocative manoeuvres. Consequently, the diagnosis of TOS is mostly done by exclusion. In challenging chronic cases of TOS, investigations should include electrodiagnostic, imaging and laboratory studies to rule out other potentially serious diagnoses, such as Paget-Schroetter syndrome, brachial plexopathy, and Pancoast tumour.

This case report describes the promising management of chronic neuropathy of the upper extremity, diagnosed as nTOS, with chiropractic treatment. Research into the diagnosis and conservative versus surgical management of TOS is warranted.

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