The enigma of neurogenic thoracic outlet syndrome following motor vehicle collisions

A. Ian Munro, MBBS
G. Duncan McPherson, MD, PhD

Accepted for publication Apr. 28, 2016

Correspondence to:
A.I. Munro
PO BOX 62010
BPO Arbutus BC V6J 1Z1
munro3676@shaw.ca

DOI: 10.1503/cjs.009814

Background: The concept of neurogenic thoracic outlet syndrome (N-TOS) including upper and lower plexus syndromes secondary to soft tissue neck injury after motor vehicle collisions (MVCs) has been contentious. We considered that analysis of objective data from this group of patients could provide insight into this controversial type of N-TOS.

Methods: During the 10-year period January 2001 through December 2010 we examined patients who had received a diagnosis of N-TOS following an MVC. We graded the principal diagnosis based on the objective data from our physical examination.

Results: In total 263 patients received a diagnosis of N-TOS during the study period. At the highest accuracy level of diagnosis there were 56 patients with ulnar entrapment syndrome (UES), 40 with carpal tunnel syndrome (CTS) and 55 with nonorganic disease (NOD), for a total of 151 (57.4%) cases in which the diagnosis of N-TOS was brought into question. The elevated arm stress test (EAST) reproduced the symptoms of UES in 33 of the 56 patients of UES (58.9%) and reproduced the symptoms of CTS in 18 of the 40 patients with CTS (45.0%).

Conclusion: There appears to be a high incidence of misdiagnosis of N-TOS following MVCs. The EAST is not a prime test for N-TOS.

Contexte : Il n'y a pas consensus sur le concept de syndrome du défilé thoraco-brachial (SDTB) neurogène comprenant des syndromes du plexus brachial inférieur et supérieur consécutifs à une blessure aux tissus mous du cou découlant d'une collision de véhicules motorisés. Nous avons pensé que l'analyse de données objectives sur les patients touchés pourrait aider à comprendre ce type controversé de SDTB neurogène.

Méthodes : Durant une période de 10 ans, soit de janvier 2001 à décembre 2010, nous avons examiné des patients ayant reçu un diagnostic de SDTB neurogène après une collision de véhicules motorisés. Nous avons coté le diagnostic principal selon les données objectives de notre examen physique.

Résultats : Au total, 263 patients ont reçu un diagnostic de SDTB neurogène durant la période à l'étude. Au degré le plus précis de diagnostic, 56 patients étaient atteints de syndrome canalaire du nerf cubital, 40, de syndrome du canal carpien et 55, de maladies non organiques, pour un total de 151 patients, ou 57,4 % des cas pour lesquels le diagnostic de SDTB neurogène avait été envisagé. La manœuvre du chandelier (test de Roos) a reproduit les symptômes du syndrome canalaire du nerf cubital chez 33 des 56 patients atteints (58,9 %) et ceux du syndrome du canal carpien chez 18 des 40 patients atteints (45,0 %).

Conclusion : Il semble y avoir une forte incidence de mauvais diagnostics de SDTB neurogène après des collisions de véhicules motorisés. Le test de Roos n'est pas un test de premier choix pour ce syndrome.

During the first half of the twentieth century the diagnosis of neurogenic thoracic outlet syndrome (N-TOS) became established under a variety of names, including scalenus anticus syndrome, and the extensive terminology has been collated by Crawford.[1] Peet and colleagues[2] are credited with introducing the term "thoracic outlet syndrome" in 1956. However, in 1944 Walshe and colleagues[3] had already stressed that the various clinical syndromes should be thought of primarily "in terms of an abnormal upper thoracic outlet." Their use of the term upper thoracic outlet initiated the confusion between the anatomic term "thoracic inlet" and the

clinical term "thoracic outlet." Today it is generally accepted that, although rare, N-TOS is due to pressure on the lower trunk of the brachial plexus, which carries the anterior primary rami of the eighth cervical spinal nerve (C8) and the first thoracic spinal nerve (T1), thus giving symptoms and signs in the dermatomes and myotomes of C8 and T1 in the upper limb.

In 1913, Wilson[4] described cases with wasting of the muscles of the thenar eminence, especially the abductor pollicis and opponens pollicis, and with sensory change along the radial border of the hand and fingers in association with a cervical rib. He discussed treatment by excision of the rib. In retrospect he was probably describing carpal tunnel syndrome (CTS). Also in 1913, Marie and Foix[5] described spontaneous compression of the median nerve in the carpal tunnel, including the autopsy findings, but this went unnoticed at the time. In 1938, Moersch[6] also described spontaneous compression of the median nerve in the carpal tunnel and warned against an incorrect diagnosis of cervical rib. Following further studies by Woltman[7] in 1941, Zachary[8] in 1945, Brain and colleagues[9] in 1947 and Kremer and colleagues[10] in 1953, there was increasing recognition of CTS, and by 1959 a widely read textbook of surgery stated, "It is now becoming recognized that many of the symptoms which in the past were ascribed to the cervical rib and the 'costoclavicular syndrome' are in reality the result of compression of the median nerve in the wrist at the site of the carpal tunnel."[11] However, in 1966 the concept of the upper plexus N-TOS was revived by Roos and Owens,[12] later supported by Urschel and Razzuk[13] in 1998 and by Sanders and colleagues[14] in 2000. As a result, the hypotheses of upper plexus and lower plexus N-TOS have been used to explain a variety of neurologic symptoms in the upper limb following motor vehicle collisions (MVCs) and alleged to be due to soft tissue injury of the neck leading to hypothetical scalene muscle spasm and compression of the brachial plexus. The existence of this form of N-TOS has been very controversial, with little objective evidence but strong opinions resulting in claims that it is either underrated or overdiagnosed.[15,16]

We have had the opportunity to carry out independent medical-legal examinations of patients with diagnoses of N-TOS secondary to an MVC. Analysis of the objective data from these examinations has provided some insight into this controversial form of N-TOS. To our knowledge, this type of objective data has not been available in the medical literature before.

Methods

We analyzed the cases of all patients referred for an independent medical opinion concerning a diagnosis of N-TOS or following an MVC and seen between Jan. 1, 2001, and Dec. 31, 2010.

Document reviews without examination of the patient were excluded. Each patient had been referred by a lawyer or an insurance company representative. It should be noted that for residents of the province of British Columbia there is mandatory MVC coverage by a single insurance agency, the Insurance Corporation of BC.

We obtained a full history, carried out a physical examination with special attention to the upper extremities, reviewed the results of investigations and reviewed the medical records that had been sent to us. The results of the physical examination were recorded on a standardized form and included the range and strength of joint movement in the upper limbs; a detailed examination of the muscles involved in entrapment neuropathies of the upper limb (especially flexor digitorum profundus, abductor digiti minimi, abductor pollicis brevis and opponens pollicis); and the provocative tests reported to be indicative of N-TOS, including the hyperabduction test and the elevated arm stress test (EAST).

The hyperabduction test was carried out with the patient seated. The arm was passively abducted in the coronal plane while the radial pulse was monitored by the examiner. The angle of elevation was based on the line of the humerus in relation to vertical when the comfortable limit of abduction had been reached. The EAST was also carried out with the patient seated; the arms were abducted to 90° in the coronal plane with the forearms vertical and palms forward. After checking the radial pulse, the examiner asked the patient to clamp and unclamp the hands for up to 3 minutes. The distribution of any increased or reproduced neurologic symptoms was noted.

Neurological diagnosis was based on the neuroanatomy of the dermatomes and myotomes for TOS and for cervical radiculopathy versus the sensory and motor supplies of peripheral nerves in CTS and ulnar entrapment syndrome (UES). We also took into account pain or tenderness at the site of entrapment, radiation of pain or parathesias from the site of entrapment and the results of provocative tests, such as the Phalen test for CTS and the elbow flexion test for UES. We reviewed the results of nerve conduction studies (NCS) and electromyography (EMG) when they were available. Because we were giving an independent medical-legal opinion we were unable to ensure receipt of appropriate NCS and EMG results in all cases. We did not use the results of provocative tests for TOS for diagnosis because they have never been scientifically validated; however, we did analyze the results of these tests.

For any nerve entrapment syndrome there is a spectrum of diagnosis from possible to certain. Therefore, a method of grading the accuracy of our diagnosis was required. The grading system we used is laid out in Table 1 and can be applied to N-TOS, UES and CTS. There were many patients in whom symptoms were nonanatomic, nonphysiologic and associated with multiple disparities on physical examination (analogous to Waddell signs in low back

examination). A grading system was devised for these cases of nonorganic disease (NOD) and is shown in Table 2. We made no attempt to diagnose the cause of the somatization or conversion disorder resulting in nonorganic disease.

Results

A total of 263 patients who had received a diagnosis of N-TOS were seen during the study period; 58 were men and 205 were women. All had been in an MVC, including 1 pedestrian, 1 motorcyclist and 1 farm tractor driver. Neck injury had occurred in 219 patients and resulted in a whiplash-associated disorder grade 1 or 2.[17] The remaining 44 (16.7%) patients, had no mention of a clinically important neck injury in the medical records early after the MVC. Therefore, injury of the scalenus anterior muscle secondary to a neck injury cannot be an essential factor in causation of N-TOS.

When seen by us, 260 patients had symptoms of tingling or numbness in 1 or both upper limbs, 1 patient reported tremors, 1 reported feelings of "electricity" and 1 had no neurologic symptoms but had received a diagnosis of TOS on the basis of a very easily occluded radial pulse. However, this was secondary to a Bankart operation for recurrent dislocation of the shoulder.

Table 1. Entrapment syndromes: grading of accuracy of diagnosis

Grade	Criteria
1. Possible	Symptoms only
2. Probable	Symptoms plus either sensory deficit or signs at entrapment site
3. Highly probable	Sensory deficit plus signs at entrapment site
4. Certain	Any of the above plus either specific motor loss or positive electrophysiology

Table 2. Nonorganic disease: grading accuracy of diagnosis

Grade	Criteria
1. Possible	Symptoms only
2. Probable	Two objective signs on physical examination
3. Highly probable	Three objective signs on physical examination
4. Certain	More than 3 objective signs or/plus known psychiatric disease

The principal diagnoses with the grading for diagnostic accuracy are given in Table 3. In no case was there tingling or numbness corresponding to the dermatomes of C8 and T1 or weakness corresponding to the myotomes of C8 and T1. Therefore, it is our view that no patient had recognizable TOS. We considered the possibility of the hypothetical "upper plexus" TOS, but the 2 patients with C6 and the 1 patient with C7 dermatome and myotome involvement were adequately explained on the basis of cervical radiculopathy with severe stenosis of the neural foramen on imaging studies. Because NOD, especially the more severe grades 3 and 4, may obscure the presence of organic disease, we cannot completely excluded the possibility of N-TOS obscured by NOD. The 14 patients whose cases were classified as miscellaneous are listed in Table 4.

Nerve conduction studies had been performed in certified laboratories, reported by certified specialists and were available for 196 patients. In a further 43 patients, the studies had been done but were never made available to us. For 24 patients no NCS were on record. For the grade 4 cases of CTS and UES the correlation of muscle weakness and positive nerve NCS is shown in Table 5. For grade 4 UES the high number of negative NCS, despite the presence of specific muscle weakness, was surprising. Analysis of the data sheets showed that inching studies were rarely performed and that the date of the NCS had preceded the date of our physical examination by 1–65 months (mean 22.5 mo). Furthermore, in 3 cases we could not find evidence that measurements had been taken across the elbow segment. In 1 patient the NCS had been done on the less affected side.

When seen by us, 24 patients had undergone surgery on 1 or both sides for N-TOS (23 had had first rib resections and 1 had had a scalenectomy), but most or all of their original symptoms had recurred. In our opinion, the correct diagnosis was UES in 9 patients, CTS in 4 patients and NOD in 11 patients, suggesting that the preoperative diagnosis had been incorrect.

Because the EAST has been reported to be the best of the tests for N-TOS,[12] we analyzed the results of this test for the grade 4 cases of UES and CTS. The EAST reproduced or increased the symptoms of UES in 58.9% of the

Table 3. Principal diagnosis and grade of accuracy

	Diagnosis; no. of cases						
Grade	TOS	UES	CTS	NOD	CRy*	Misc	Total cases; no. (%)
1	0	12	8	4	—	—	24 (9.1)
2	0	17	13	14	—	—	44 (16.7)
3	0	11	6	10	—	—	27 (10.3)
4	0	56	40	55	—	—	151 (57.4)
NG	—	—	—	—	3	14	17 (6.5)
Total, no. (%)	0	96 (36.5)	67 (25.5)	83 (31.6)	3 (1.1)	14 (5.3)	263 (100)

CRy = cervical radiculopathy; CTS = carpal tunnel syndrome; Misc = miscellaneous; NG = not graded; NOD = nonorganic disease; TOS = thoracic outlet syndrome; UES = ulnar entrapment syndrome.

*Sixth cervical spine nerve 6 (C6): $n = 2$; C7: $n = 1$.

grade 4 UES cases and reproduced the symptoms of CTS in 45.0% of the grade 4 CTS cases. Therefore, the EAST is not a reliable test for TOS.

In 59 patients there was significant disparity between the active and passive ranges of shoulder abduction on 1 or both sides associated with NOD. Therefore these cases were excluded from the analysis of the results of the hyperabduction test. Among the remaining 204 patients the hyperabduction test occluded the radial pulse on 1 or both sides in 59 cases (39%).

Among the 56 patients with grade 4 UES, 17 (30.4%) had a sensory deficit that included the ulnar 2 and a half digits compared with the normal expected incidence of 15%–20% for this variant. From review of the medical-legal reports, some physicians did not recognize this variant and appeared to use it to make a diagnosis of TOS, thus inflating the incidence to 30.4% in our series.

Discussion

Although the present series can be criticized for having been selected by lawyers and insurance company representatives, the original diagnosis had been made by a physician or surgeon.

Until now, to our knowledge, the only attempt to obtain objective data about N-TOS was a study by Cherington and Cherington[18] that analyzed payment agencies and N-TOS surgery in Colorado, USA. The authors concluded that no surgery had been carried out in patients who could not pay. The study questioned "whether surgery should be done at all for a condition that is vague and controversial."

Table 4. Micellaneous diagnoses

Diagnosis	No.
Raynaud	4
Occupational neck and shoulder pain	4
Minor stroke	2
Childhood head injury	1
Bicipital tendinitis	1
Bankart operation (for recurrent shoulder dislocation)	1
Lateral palmar digital nerve injury	1

Table 5. Positive NCS and specific muscle weakness in grade 4 UES and CTS

Muscle weakness	NCS	UES	CTS
Specific muscle weakness absent	NCS +	14	29
Specific muscle weakness present	NCS +	6	6
	NCS –	26	2
	NCS – or ND	10	3
Total		56	40

CTS = carpal tunnel syndrome; NCS = nerve conduction studies; ND = not done; UES = ulnar entrapment syndrome.

Eisen[19] studied the use of electrophysiology in the early diagnosis of UES by examining healthy controls and patients with mild UES and severe UES. There were 34 patients with severe UES, all of whom had "clear-cut evidence of an ulnar nerve compression at the elbow" and muscle atrophy, weakness and appropriate sensory deficit specific for UES. These 34 cases approximate to, but were probably more severe than, our grade 4 cases because not all our cases had muscle atrophy. Using Eisen's data, 21.2%– 61.6% of his NCS were normal in cases of severe UES. Thus, the presence of normal NCS may not exclude a grade 4 UES. This is in addition to the other factors contributing to normal NCS in patients with severe UES.

In our series there were 151 diagnoses of grade 4 UES, CTS and NOD. Therefore, it is probable that the diagnosis of N-TOS was incorrect in 57.4%, and it is probable that there were more cases of incorrect diagnosis among the remaining 42.6%. In our opinion, the cases of failed surgery were due to UES in 9 patients, CTS in 4 patients and NOD in 11 patients, giving further support to the suggestion that there is a high level of misdiagnosis. Our results also support MacKinnon's comments in the discussion on the study by Urschel and Razzuk[13] stating that her own group reserves surgical intervention for the unusual patient who cannot be treated for CTS or UES.

Reliance on the Roos elevated arm stress test or EAST[12] as a specific test for N-TOS contributed to the misdiagnoses. Our results show that the EAST was positive for the peripheral neuropathy in about 45% of the grade 4 cases of UES and nearly 60% of the grade 4 cases of CTS. In 1985, Costigan and Wilbourne[20] reported that the EAST produced symptoms in the fingers in 19% of patients with known CTS but none in the asymptomatic controls. The higher incidence in our group was probably due to reliance on the EAST as a specific test for N-TOS by the physician making the original diagnosis of N-TOS. The incidence of a positive EAST in patients with a primary diagnosis of UES is unknown but, like CTS, is probably less than the incidence in this series because of reliance on the EAST as a specific test for N-TOS before entering this study.

In 1941, Wright[21] showed that the radial artery pulse can be occluded in more than 80% of the healthy population with the shoulder or shoulder girdle in more extreme positions and commented that "hyperabduction is today being widely but erroneously used as a test for the scalenus anticus syndrome." Therefore, we disregarded vascular laboratory studies that reported N-TOS on the basis of arterial pulse occlusion. Our result for easy radial artery pulse occlusion was 39%, whereas Wright's most easily occluded cases were 32%.

Other factors possibly contributing to misdiagnosis were lack of knowledge of the neuroanatomy of the upper limb and reliance on a diagnosis of "double crush syndrome" to explain any evidence of a peripheral neuropathy. These possibilities were not analyzed in the present study.

Finally, combining the diagnoses in Table 3, Table 4 gives our experience in the differential diagnosis of TOS during the 10 years analyzed.

Conclusion

Our results suggest that there is a high incidence of error in the diagnosis of N-TOS in patients presenting for medical-legal insurance examinations following MVCs. Recurrence of symptoms following first rib resection may be associated with an incorrect preoperative diagnosis. In our experience the EAST is not specific for TOS because it can be positive for CTS or UES. Our results should be confirmed by others.

Acknowledgements: The authors thank their teachers of anatomy at Western University, Ontario, and Kings College, London, without whom this paper would not have been possible, and Dr. W.R.E. Jamieson for helpful advice.

Affiliations: From the Department of Surgery, University of British Columbia, Retired (Munro); and the Department of Orthopedics, University of British Columbia, Retired (McPherson).

Competing interests: None declared.

Contributors: Both authors designed the study. A.I. Munro acquired and analyzed the data and wrote the article, which both authors reviewed and approved for publication.

References

1. Crawford FA. Thoracic outlet syndrome. *Surg Clin North Am* 1980; 60:947-56.
2. Peet RM, Hendrikson JD, Anderson TP, et al. Thoracic outlet syndrome: evaluation of a therapeutic exercise program. *Proc Staff Meet Mayo Clin* 1956;31:281-7.
3. Walshe FMR, Jackson H, Wynburn-Mason R. On some pressure effects associated with cervical and with rudimentary and 'normal' first ribs, and the factors entering into their causation. *Brain* 1944; 67:141-77.
4. Wilson SAK. Some points in the symptopathology of cervical rib, with especial reference to muscular wasting. *Proc Columbia Hospital Soc Med* 1913;6:133-41.
5. Marie P, Foix MC. Atrophie isolee de l'eminence thenar d'origine nevritique: role du ligament annulaire du carpe dans la pathogenie de la lesion. *Rev Neurol (Paris)* 1913;26:647-9.
6. Moesch FP. Median thenar neuritis. Proc. Staff Meetings Mayo Clinic, 1938; 220-222.
7. Woltman HW. Neuritis associated with acromegaly. *Arch Neurol Psychiatry* 1941;45:680-2.
8. Zachary RB. Thenar palsy due to compression of the median nerve in the carpal tunnel. *Surg Gynec Obst* 1945;81:213-17.
9. Brain WR, Wright AD, Wilkinson M. Spontaneous compression of both median nerves in the carpal tunnel. *Lancet* 1947;1:277-82.
10. Kremer M, Gilliat RW, Golding JSR, et al. Acroparaesthesiae in the carpal tunnel syndrome. *Lancet* 1953;265:590-5.
11. Bailey M, Love M, Charnley J, et al. *Short practice of surgery*. H.K. Lewis & Co. Ltd., London, 1959: p. 1302.
12. Roos DB, Owens JC. Thoracic outlet syndrome. *Arch Surg* 1966; 93:71-4.
13. Urschel HC, Razzuk MA. Neurovascular compression in the thoracic outlet. Changing management over 50 years. *Ann Surg* 1998;228:609-17.
14. Sanders RJ, Cooper MA, Hammond SL. Neurogenic thoracic outlet syndrome. In: *Vascular Surgery*; W. B. Saunders Company, 2000:p. 1184-1200.
15. Roos DB. The thoracic outlet syndrome is under-rated. *Arch Neurol* 1990;47:327-8.
16. Wilbourne AJ. The thoracic outlet syndrome is over-diagnosed. *Arch Neurol* 1990;47:328-30.
17. Quebec Task Force on Whiplash-Associated Disorders [WAD]; Section 2, p5-6. Ville de Quebec. Que. Societe d'Assurance Automobile du Quebec, 1995.
18. Cherington M, Cherington C. Thoracic outlet syndrome: reimbursement patterns and patient profiles. *Neurology* 1992;42:943-5.
19. Eisen A. Early diagnosis of ulnar nerve palsy. An electrophysiologic study. *Neurology* 1974;24:256-62.
20. Costigan DA, Wilbourne AJ. The elevated arm stress test in the diagnosis of the thoracic outlet syndrome. *Neurology* 1985;35(suppl. 1):74-5.
21. Wright IS. The neurovascular syndrome produced by hyperabduction of the arms. *Am. Ht. J.* 1945;29:1-19.