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TRUE NEUROGENIC THORACIC OUTLET SYNDROME IN A COMPETITIVE SWIMMER

A case report of this rare association

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ABSTRACT - True neurogenic thoracic outlet syndrome (TOS) is an uncommon disorder despite of be a frequent reason for referral to the EMG laboratories. We describe the second case in the literature of true TOS in a competitive swimmer with progressive weakness and severe atrophy of the left thenar eminence. EMG showed lower trunk plexopathy. X-ray and MRI of the cervical spine and brachial plexus were normal. Surgical exploration evidenced the lower trunk retracted and pulled by a fibrous band. It was excised and neurolysis of the plexus was done with gradual improvement of function. We discuss the possible pathophysiology of this association.

KEY WORDS: thoracic outlet syndrome, swimming, brachial plexopathy.

Síndrome do desfiladeiro torácico verdadeiro em um nadador competitivo: relato de caso desta rara associação

RESUMO - A síndrome do desfiladeiro torácico verdadeiro é condição incomum, apesar de ser uma razão freqüente de encaminhamento aos laboratórios de eletroneuromiografia. Nós descrevemos o segundo caso na literatura desta doença rara em um nadador competitivo com fraqueza e atrofia severa da eminência tenar esquerda. A ENMG mostrou plexopatia do tronco inferior. O RX e as ressonâncias magnéticas da coluna cervical e do plexo braquial foram normais. Exploração cirúrgica mostrou o tronco inferior tracionado por uma banda fibrosa. Ela foi ressecada e procedeu-se a neurólise do plexo com melhora gradual da função. Nós discutimos a possível fisiopatologia desta associação.

PALAVRAS-CHAVE: síndrome do desfiladeiro torácico, natação, plexopatia braquial.

Thoracic outlet syndrome (TOS) is a term coined by Peet et al in 1956 to a wide variety of symptoms originated by compression of the neurovascular bundle at the transition between the neck and axilla[1]. There are very few diseases so controversial in the medical literature such as TOS[2,3]. Based on these controversies, the TOS can be divided in vascular and neurogenic[3]. The vascular TOS can be subdivided in arterial and venous according to the compression of the subclavian artery or vein respectively. They result in ischemia of the digits and hand or swelling of the arm and represent only 5% of the patients with TOS[4]. The neurogenic TOS represent the rest 95% and can be subdivided in true and disputed.

Following the ongoing debate in regard to the entity of TOS over the past two decades, a consensus has emerged that classic (true) neurogenic TOS is uncommon and is usually caused by compression of the lower trunk of the brachial plexus due to a cervical rib or band and enlarged scalenus muscles[5-7]. This compression results in arm pain, numbness of the inner surface of the hand and forearm, and characteristic wasting and weakness of the thenar and intrinsic hand muscles. Distinct electrophysiological abnormalities (low compound muscle action potential in the thenar and intrinsic muscles, abnormal sensory nerve conduction in the ulnar nerve, prolonged F-wave latency in the ulnar nerve, and abnormal medial antebrachial cutaneous sensory nerve conduction) are noted in classic neurogenic TOS[8-10].

TOS has been described in aquatic athletes including competitive swimmers, divers, water polo players, and synchronized swimmers[11,12]. Most of these cases did not have neurological deficits, representing non-

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classic or disputed TOS. There has been just one reported case of classic (true) neurogenic TOS in competitive swimmers[13], and we are reporting another such case.

CASE

Seven weeks prior to the initial evaluation, a 17-year-old right-handed male suddenly developed dragging of the fifth finger of left hand followed by progressive weakness and atrophy involving the whole left hand, after participation in competitive swimming. He denied any sensory symptoms and reported no back, arm or hand pain. There was no recent history of acute trauma, immunization, or flu-like symptoms. He had been a competitive freestyle swimmer for several years. Family history was positive for diabetes and coronary artery disease.

The general physical examination was normal. On neurological examination the patient was alert and oriented, with normal cognitive function and cranial nerves. No Horner's sign was seen. There was weakness (grade 3-4 on MRC scale) of thenar, hypothenar and intrinsic muscles of the left hand. Moderate atrophy in the left thenar, hypothenar and interosseous muscles was noted, giving an impression of smaller hand (Figure). Mild atrophy of the left forearm flexor muscles was noted as well. Deep tendon reflexes were normal and symmetrical. Sensation was intact to pinprick, light touch, temperature and proprioception. There was evidence of decreased vibratory sensation in the left fingers. At the supraclavicular fossa, no palpable mass or tender spot was noted, but inconsistent Tinel's sign with radiating sensation to the digit V was present.

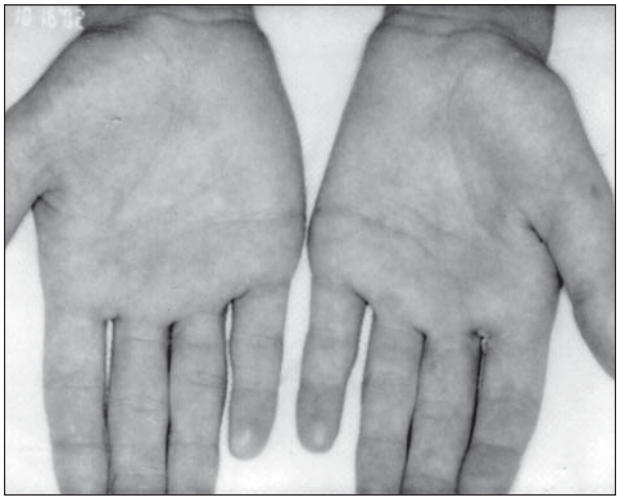

Figure. Atrophy of left thenar and hypothenar muscles.

X-rays of the cervical spine and thorax did not show any cervical rib. MRI scans of the brain and left brachial plexus were unremarkable, and MRI of the cervical spine showed insignificant left paraspinal disc protrusion at the C5-C6 level without spinal canal or foraminal compromise.

The nerve conduction study showed prolonged F-wave latencies in the left median and ulnar motor nerves, prolonged terminal latency and low CMAP amplitude in the left ulnar nerve, and normal NCS in the left ulnar and median sensory nerves (Table 1). No sensory compound nerve action potential (CNAP) was recorded from the left medial antebrachial cutaneous nerve. The needle EMG study re-

Table 1. Nerve conduction data.

	11/16/01		4/8/03 (PO 10 m)		Normal	
	L/NCV*	Amp**	L/NCV*	Amp**	L/NCV*	Amp**
Sensory nerve conduction						
Media	46.4	27.0	46.4	31.0	41.3	10
Ulnar	44.0	18.0	44.2	19.8	39.3	10
Medial ABC	**NP*** **		48.1	**3.3**	41.7	10
Mixed nerve conduction						
Median: Wrist-elbow	55.6	64.0	54.5	51.0	49.4	10
Elbow-axilla	58.1	89.0	57.7	64.0	53.4	10
Ulnar: Wrist-elbow	61.0	18.0	53.4	40.0	47.5	10
Elbow-axilla	69.0	56.0	52.4	42.6	48.1	10
Motor nerve conduction						
Median: Terminal latency	2.9	6.6	2.9	11.8	3.6	5
Elbow-wrist	60.0	6.4	54.1	11.3	50.0	5
Axilla-elbow	56.3	5.6	54.2	11.2	56.0	5
F-wave	**32.3**		**29.8**		29.7	
Ulnar: Terminal latency	**3.0**	5.0	**3.3**	**4.6**	2.5	5
Elbow-wrist	58.9	**4.5**	51.4	**4.0**	50.6	5
Axilla-elbow	81.0	**4.5**	52.4	**3.7**	52.3	5
F-wave	**36.3**		**34.2**		30.3	

*L/NCV, Latency (msec)/ NCV (m/s); **Amplitude of compound nerve action potential (μV) for sensory and mixed nerve conduction and amplitude of compound muscle action potential (mV) for motor nerve conduction; ***Normal side: 52.2 m/s in NCV and 12 μV in amplitude. PO, post-operative; ABC, antebrachial cutaneous; NP, no potential. Bold numbers represent abnormal values.

Table 2. Cases with thoracic outlet syndrome among swimmers.

Cases	Swimmer	Symptoms	Findings	Cause	Treatment
Non classic (disputed) neurogenic TOS					
Frankel (17)					
Case 1	Free style swimmer	Numbness over the right forearm and hand.	Hypoesthesia in the ulnar territory. Scalenus mass, Tinel sign.	Hypertrophied scalenus anticus muscle.	Scalenotomy. Improved.
Case 2	Varsity swimmer	Aching in the right forearm and hand ulnar aspect.	Hypoesthesia in the ulnar territory. Tinel sign & tender scalenus muscle.	Hypertrophied scalenus anticus muscle.	Scalenotomy. Improved.
Richardson (11)					
Case 1	Varsity swimmer	Shoulder numbness and tingling radiating to the IV and V fingers.	No neurological deficit.	No cervical rib.	First rib removal. Improved.
Case 2	Waterpolo player	Shoulder pain radiating to the forearm, the IV and V fingers.	No neurological deficit.	No cervical rib.	First rib removal. Improved.
Case 3	Varsity swimmer	Shoulder pain radiating down to to the IV and V fingers.	No neurological deficit.	No cervical rib.	No surgery.
Classic (true) neurogenic TOS					
Katirji (13)					
Case 1	Free style/ butterfly swimmer	Weakness and atrophy of the right hand. Hypoesthesia in the ulnar aspect of forearm and hand.	Atrophy and weakness of thenar and intrinsic hand muscles. Abnormal EMG/NCS.	Hypertrophied scalenus anticus muscle with band.	Muscle & band sectioned. No improvement.
Oh (*)					
Case 1	Free style swimmer	Weakness and atrophy of the left hand.	Atrophy and weakness of thenar and intrinsic hand muscles; Tinel sign ± impaired vibration on fingers. Abnormal EMG/NCS.	Fibrous band.	Band section. Improved.

*The present case.

vealed positive sharp waves and fibrillations with high-amplitude, long-duration motor unit potentials in the left first dorsal interosseous and abductor pollicis brevis muscles. The left biceps, deltoid, triceps and paraspinal muscles showed normal EMG findings.

Over the next three months, there was minimal improvement in strength in the intrinsic hand muscles after the patient stopped competitive swimming. In view of such minimal improvement, surgical exploration was recommended for classic neurogenic TOS. It showed entrapment of the lower trunk of the brachial plexus by a pleural band causing retraction of this segment over the middle and upper trunks. The abnormal band was excised and neurolysis of the plexus was done to avoid further entrapment and scarring around the plexus. There was mild improvement in the strength of intrinsic hand muscles (finger-spreading increased from 3 to 4 by MRC scale and hand dynamometry from 12 kg to 17 kg), but no improvement was noted in muscle atrophy over a 2-year follow-up period. Nerve conduction study 10 months after the surgery showed definite improvement: the CMAP amplitude from the abductor pollicis brevis muscle was increased from 6.6 to 11.6 mV and the sensory CNAP, though the amplitude was low, was obtained in the left medial antebrachial cutaneous nerve (Table 1).

DISCUSSION

Our patient had progressive muscle weakness and wasting in the left hand, which are the hallmarks of classic (true) neurogenic TOS. There was no history of pain or sensory complaints as typically seen in non-neurogenic TOS. Imaging work-up was not able to demonstrate any cervical rib or enlarged scalenus muscle. Nerve conduction studies showed almost all the typical electrophysiological abnormalities of neurogenic TOS (low CMAP amplitude, more often in the median than in the ulnar nerve, prolonged ulnar F-wave latency, abnormal ulnar sensory nerve conduction, and absent medial antebrachial cutaneous sensory nerve conduction)[9,10]. Low CMAP amplitude in the median nerve was not evident until the post-operative second NCS which showed almost doubling of the CMAP amplitude. One exception was the normal sensory nerve conduction of ulnar nerve. In recent years, the medial antebrachial cutaneous sensory CNAP seems to be the most sensitive nerve conduction parameter to confirm classic neurogenic TOS[14,15]. It is particularly helpful when the ulnar sensory CNAP is normal and TOS is clinically suspected. Kothari et al. believe that the medial antebrachial cutaneous is more sensitive than ulnar sensory nerve conduction study because the former carries predominantly T1 fibers instead of C8 fibers carried by the ulnar sensory nerve[14]. It is widely accepted that the T1 fibers are

predominantly involved in true neurogenic TOS. Anatomical preference would explain why the abductor pollicis brevis muscle is usually more atrophied than the first dorsal interosseous and the median CMAP is more severely affected than the ulnar CMAP.

Shoulder pain has always been a common complaint of athletes who use their arms extensively for sports[12]. Swimmers are the most frequently affected group of competitive athletes: 42% of America's best swimmers have "swimmer's shoulder"[11]. Swimmer's shoulder is thought to be an impingement syndrome due to chronic irritation of the humeral head and rotator cuff on the coracoacromial arch during abduction of the shoulder[11,16]. This is said to be due to the controlled, repetitive power motion at the very extreme of abduction and external rotation of the shoulder which is required in freestyle, butterfly, and backstroke swimming[11]. TOS has been included in the differential diagnosis of shoulder pain in the swimmer[11]. Seven cases of TOS including our case have been reported among swimmers (Table 2). Five cases were non-classic or disputed neurogenic TOS due to lack of any objective neurological deficit. Two patients of Frankel et. al. had hypertrophied scalenus anticus muscle and have improved after scalenotomy[17]. Two of three patients of Richards et. al had symptomatic relief with removal of the first rib and resumed competitive swimming[11]. Only one patient of the literature had classic neurogenic TOS[13]. During the surgical exploration there was no cervical rib but a hypertrophied scalenus anticus muscle with a fibrous band. There was no improvement after section of the band and neurolysis[13]. There is also a report of one case of "effort thrombosis" of the subclavian vein in a competitive swimmer[18], without any evidence of neurogenic TOS.

The mechanism of classic neurogenic TOS in the swimmer was thought to be due to hypertrophy of the scalenus anticus muscle as an expression of overdevelopment of the neck and shoulder muscles, which results from many years of training in this sport[17]. Hypertrophy of the scalenus anticus muscle was also seen in Katirji and Hardy's case[13]. However, they also described a fibrous band within the scalenus anticus muscle, which could be the cause of the entrapment. In our patient, there was no hypertrophic scalenus muscle but a fibrous band compressing the lower trunk of the brachial plexus was present. We believe that congenital fibrous bands were the cause of classic neurogenic TOS in these two patients, and that competitive swimming aggravated or precipitated TOS symptoms.

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