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Thoracic outlet syndrome: definition, aetiological factors, diagnosis, management and occupational impact

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Clinical review

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Abstract

Introduction

Thoracic outlet syndrome is a controversial cause of neck and shoulder pain due to complex mechanisms involving muscular dysfunction and nerve compression. Although management of thoracic outlet syndrome must be based on a multidisciplinary approach, physicians and occupational therapist should be familiar with the principles of diagnosis and treatment.

Method, results and conclusion

The purpose of this article is to review the definitions, diagnosis and management of this syndrome. A particular emphasis was described on the links between the workplace and the individual in the pathogenesis, prevalence in the workforce and the course of this disease.

Key words: Thoracic outlet syndrome, rehabilitation, diagnosis, occupational disease

Introduction

Shoulder disorders, which include unspecific shoulder pain and specific disorders, are commonly diagnosed in primary care and often lead to prolonged disability. Their 12-month prevalence in the population of working age range between 7 to 47% for shoulder pain, depending on the population studied and the definition used [1]. The impacts for workers are important in industry such as in office, especially for chronic shoulder pain [2].

Although neck and arm pain is a frequent presenting complaint in the general population, an unusual and controversial cause must sometimes be considered [3-4]: thoracic outlet syndrome (TOS), as this frequently complex disease is difficult to diagnose. A diagnosis of TOS can be rapidly considered by physicians with the clinical features suggestive of TOS and the risk factors and occupational situations associated with it. Considering multiple aspects of the outcome including pain, general physical function and work, rapid referral to specialized multidisciplinary units may then allow more effective management of this disease.

The purpose of this study is to review the definitions of TOS, the known risk factors, diagnostic criteria, and management. The links between TOS and the work environment will be described in particular detail on prevalence in the workforce, occupational risk factors, and work prognostic factors (including the key message for clinicians and rehabilitation professionals).

Definitions

Thoracic outlet syndrome covers a wide range of manifestations due to compression of nerves and vessels during their passage through the cervicothoracobrachial region. Various forms of TOS are distinguished: vascular forms (arterial or venous) which raise few diagnostic problems [3], and “neurological” forms, which are by far the most frequent as they represent more than 95% of all cases of TOS [5, 6]. The “neurological forms” are classified in the "true" neurological form associated with neurological deficits (mostly muscular atrophy), and painful neurological forms (with no objective neurological deficit). These painful forms are very frequent, especially when patients are systematically screened for these symptoms. The existence of these forms of TOS remains controversial in part because muscular and neurological manifestations are strongly interrelated. Clinical experience suggests that the main triggering mechanism is more often a muscular dysfunction in the cervicoscapular region than primitive nerve compression. It is directly responsible for cervicoscapular symptoms (pain and discomfort) and sometimes for referred scapulobrachial and facial pain. In parallel, shortened muscles (mainly scalene muscles) and cervicoscapular muscles imbalance may lead to intermittent nerve compression and/or tension on brachial plexus in the thoracic outlet resulting in proximal pain and producing pain and discomfort in the upper limb. The neurological involvement accounts for most of the distal symptoms, but the controversy concerning the reality of TOS is essentially due to the absence of objective criteria to confirm the diagnosis (no neurological weakness and normal neurophysiological examination). Despite considered as “debatable” for some authors, several arguments support the reality of this syndrome, such as the influence of TOS on the results of treatment of carpal tunnel and cubital tunnel syndromes [7,8]. This problem is further complicated by the frequent concomitant presence of other neuromuscular diseases of the upper limb, which can be secondary to TOS or, on the contrary, may precede and predispose to the development of TOS

[7, 8], in which case TOS is often masked by the concomitant disease. “Neurological” forms of TOS can be subdivided into primary forms in which features of TOS may remain isolated or may be complicated by underlying neuromuscular disorders, and forms secondary to a more distal disease (neuromuscular or joint disease), always responsible for complex clinical features.

Aetiological forms

Painful forms of TOS can be due to four main causes, sometimes interrelated.

1/ Congenital abnormalities are often reported and can be associated with traumatic or functional causes. Bone anomalies (cervical rib, prolonged transverse process), fibrous anomalies (transversocostal, costocostal, etc.), or muscular anomalies (scalenus anticus muscle, sickle-shaped scalenus medius, etc.) are more frequent in patients who develop TOS [5]. Bone anomalies are well known, but 2/3 of the abnormalities detected at operation are fibromuscular [5] and the majority of bone anomalies do not cause TOS [9, 10]. These anomalies are part of a real local and regional “dysplasia” constituting only one of numerous predisposing factors, associated with a morphotype composed of narrow, drooping shoulders.

2/ Post-traumatic causes, either due to isolated trauma or repeated trauma, account for up to 2/3 of cases in some series [6, 11]. Post-traumatic TOS due to soft tissue injury raises medicolegal and often management problems. These forms are related to neck and shoulder trauma, particularly “whiplash” injuries, or sometimes upper limb trauma. Injuries to scalene muscles and their subsequent fibrosis are implicated in this process [11, 12]. Diagnostic criteria of post-traumatic TOS are the pathogenic mechanism and the onset of symptoms within the first two years.

3/ “Functional” acquired causes are the most controversial, although probably the most frequent. In this group, upper limb dysfunction or a muscle imbalance of the neck and shoulder region is considered to be responsible [11, 13-15]. Two main features are mainly associated with "functional" acquired causes: "hypertrophic" muscle morphotype of the cervicospinal region and "dropped" scapular morphotype (leading in some patients to a dysfunction of the normal scalenus anterior muscle). Muscles of the thoracic outlet are also accessory respiratory muscles capable of prolonged tonic contractions due to their high percentage of type I muscle fibres [16]. Chronic stimulation of these muscles has also been shown to increase the percentage of type I fibres. Machleder showed that a normal scalenus anterior muscle contains 70% of type I fibres versus 85% in the case of TOS [16]. The factors involved in the pathogenesis of these disorders include overuse and physical and mental stress phenomena, frequently associated with unfavourable psychosocial factors [17].

Neck and shoulder symptoms are reported by 45% of subjects in certain occupations [18] and are related to repetitive movements and certain working positions, particularly in occupations requiring use of the arms in elevation (barbers, switchboard operators, assembly lines, etc.), with the head or shoulders flexed anteriorly (secretaries, computer operators, etc.) [14, 15, 19].

4/ Other acquired causes are rare but must be systematically considered: tumours [20], hyperostosis, osteomyelitis, etc. The diagnosis is based on clinical examination and medical imaging (CT and MRI).

Diagnosis

Symptoms

Vascular forms of TOS can be either venous or arterial. Venous compression can be responsible for oedema or cyanosis of the upper limb. It can also present suddenly in the form

of phlebitis occurring after varying degrees of effort. The patient may sometimes only present at the stage of sequelae with a thoracic collateral venous circulation. Arterial signs consist of either ischaemia on effort of the upper limb or positional vasomotor disorders. Signs of vertebrobasilar insufficiency, due to compression of the origin of the vertebral artery, or Raynaud's phenomenon may be observed [4]. The diagnosis of TOS is relatively simple in the presence of vascular symptoms in the upper limb when the arms are raised, but vascular forms are rare, as venous forms represent 2 to 3% of TOS and arterial forms represent about 1% [6]. However, the presence of vascular signs may help to guide the diagnosis in the presence of a predominantly neurological form of TOS.

TOS with a muscular atrophy is exceptional and remains asymptomatic for a very long time. Very rarely, the patient may present with progressive atrophy of the intrinsic muscles of the hand, always starting with the thenar muscles and gradually spreading to the interosseous and hypothenar muscles [21]. In these forms of TOS associated with muscular atrophy, pain and paraesthesiae are often moderate or may even be absent. At an advanced stage, treatment may still be able to relieve pain, but the possibilities of motor recovery are very limited and sequelae are frequent [3, 10, 21, 22].

Painful neurological forms of TOS account for 97% of all cases of TOS according to Roos [6]. They are typically responsible for symptoms in the C8-T1 distribution (medial aspect of the arm, ulnar border of the forearm and hand), but the C7 nerve root and sometimes the superior trunk of the brachial plexus (C5-C6) may be responsible. Clearly systematized symptoms are rarely present. It has been reported that the 3 most disturbing preoperative symptoms are pain at rest (87% of cases), feeling of numbness (66% of cases) and decreased strength (55% of cases). In practice, the patient often reports vague, poorly defined, and inconsistent symptoms, but clinical interview often reveals difficulties during activities requiring elevation of the arms (hanging up the washing, brushing one's hair, etc.). Functional

impairment and pain related to carrying heavy loads are frequent but less specific. Pain of the neck and shoulder region is at least partly due to muscle imbalance but can sometimes be due to a proximal form of TOS (C5-C6). Decompensation fairly frequently occurs after a change of job or in a context of carpal tunnel syndrome (double crush syndrome) [23, 24]. These patients present with complex upper limb pain [25]. Carpal tunnel symptoms appear rapidly [26] due to pre-existing irritation of the nerve fibres by TOS [23, 24, 27]. However, the pathophysiology of the association between upper-limb distal nerve entrapment and TOS is complex, and may not be only on irritation of nerve fibres. For instance, median nerve sensory fibres do not travel with the C8 fibres that are being hypothesized as the site of the double crush. Therefore, other mechanisms could be considered in relationship to increase median or ulnar nerve pressure and scalene muscle activity [28], or a hypothesis of centralization of pain [29]. The associated TOS must be identified, as it can be responsible for persistent symptoms after treatment of carpal or ulnar tunnel syndrome [7, 8, 30].

Apart from double crush syndrome, other secondary painful diseases may be associated, such as epicondylar pain secondary to medial or lateral insertion tendinitis. However, referred pain is not always easy to distinguish from a possible associated tendinitis (medial aspect of the elbow [19, 31]), which also raises the problem of the real (or at least the initial) cause of the pain [31]. Nevertheless, some of these forms of medial or lateral epicondylitis may resolve in response to rehabilitation for TOS.

Physical examination

At first sight, the physician may observe that the patient's upper limb is relatively immobile. The shoulder can be lowered and protracted [32]. Muscles of the scapular region and the

scalene muscles may be contracted [33, 34]. The supraclavicular fossae may appear to be filled due to a cervical rib.

The physician must look for cyanosis or oedema of the upper limb or even a thoracic collateral venous circulation. The presence of a supraclavicular murmur is also an indication for complementary vascular investigations. Vascular tests (Adson, Wright, etc.) are of little value in neurological forms of TOS [6]: the presence of dynamic compression is not synonymous with TOS, as it is detected in more than 50% of the general population [5, 6]. Positive vascular tests can only help to guide the diagnosis, but do not constitute formal diagnostic criteria *per se*.

Signs of neurological deficit, essentially motor weakness (intrinsic muscles), must be systematically investigated in the hand. We have seen several patients followed for many years for “cervicobrachial neuralgia”, in whom the diagnosis of TOS was only proposed at the stage of atrophy of all intrinsic muscles of the hand, resembling an Aran-Duchenne hand [22]. However, objective clinical signs of muscular weakness are usually absent.

Stress tests are therefore particularly valuable. In the Roos stress test, the patient positions the shoulders in 90° of abduction with the elbows flexed to 90° and repeatedly opens and closes the hand [6]. This test has a fundamental diagnostic value provided it triggers the symptoms spontaneously experienced by the patient in less than one minute [15]. Similarly, the presence of a supraclavicular positive Tinel sign has a major diagnostic value, but is less often present. Elvey’s test modified by Sanders (90° of abduction/external rotation of the upper limb, wrist in extension, then the head is tilted to the contralateral side [35]), may also be useful. Finally, Morley’s sign (tenderness in the supraclavicular fossa) may have a diagnostic value when it is clearly asymmetrical and especially when it triggers the patient’s usual, more distal pain [36]. These tests are quite sensitive but poorly specific. However, association of positive tests increased the probability of the TOS.

Complementary investigations

None of the various complementary investigations are truly useful for the diagnosis of painful forms of TOS. They are especially useful in rare vascular forms, in forms associated with neurological deficits, and prior to surgery. Standard radiographs, centered on the cervicothoracic spine, can demonstrate a cervical rib or a prolonged C7 transverse process. Such an abnormality, on its own, is not sufficient to confirm the diagnosis of TOS, as only 5 to 10% of cervical ribs are associated with TOS [3, 9]. Some authors consider that CT scan can identify abnormalities in 30 to 60% of cases [15], but once again the presence of abnormalities is not synonymous with TOS [9]. MRI is of limited value for the diagnosis of TOS [26]. However, these examinations are essential when a tumour is suspected.

Static and dynamic vascular examinations (Doppler ultrasound and angiography) are only useful in the presence of vascular clinical signs. The presence of arterial stenosis or even post-stenotic aneurysm is a formal indication for surgery. Vascular examinations are of limited diagnostic value in isolated neurological forms [37]. It must be remembered that the presence of dynamic vascular compression is not synonymous with TOS.

Electroneuromyographic signs in favour of the diagnosis of TOS are: signs of chronic partial denervation in intrinsic muscles of the hand, decreased amplitude of sensory evoked potentials of the ulnar nerve and motor evoked potentials of the median nerve. Electroneuromyography (ENMG) only reveals abnormalities in severe cases and conduction velocities are only decreased in the case of permanent nerve compression. A reduction of the action potential in the territory of the cutaneous nerve of the forearm may be an earlier sign.

ENMG is usually normal. Many authors consider that the diagnosis of TOS can be raised before the appearance of signs of intrinsic muscle denervation [6, 15]. At an advanced stage, treatment may still be able to relieve pain, but the possibilities of motor recovery are very

limited and sequelae are frequent [3, 10, 22, 23]. The main role of ENMG is to detect an associated upper limb distal tunnel syndrome, like carpal tunnel syndrome or ulnar nerve at the elbow. Somatosensory evoked potentials are probably of no value for the diagnosis of TOS.

Thoracic outlet symptoms have generally been present for several years and are more or less well tolerated by the patient. Symptoms tend to be exacerbated by unusual activities or periods of stress.

Secondary TOS can also be observed in combination with any painful disease of the upper limb, especially when symptoms are longstanding and/or disabling, or when they occur in an unfavourable socioeconomic context [17, 32]. The patient develops more or less adapted compensations with secondary dysfunction of the whole upper limb. In this case, the secondary imbalance of neck and shoulder muscles is responsible for TOS, which in turn exacerbates the symptoms and can be responsible for chronic disorders. Epicondylar pain associated with TOS shows a poorer response to treatment [26].

Differential diagnosis

The absence of clinical signs of muscular weakness and usually ENMG signs requires a very rigorous diagnostic approach. At this stage, TOS must be a diagnosis of exclusion.

The diagnosis of vascular forms is usually fairly straightforward, as most of the symptoms and signs are suggestive of TOS. On the other hand, Raynaud's phenomenon is rarely related to TOS (3 to 5% of cases) and other aetiologies should be investigated.

The differential diagnosis of upper limb pain includes: other causes of brachial plexus pain, cervical pain, tunnel syndromes, degenerative disease of the upper limb, and non-compressive central and peripheral neurological disease.

The other causes of brachial plexus pain to be systematically considered are: Pancoast syndrome, radiation-induced brachial plexopathy and Parsonage-Turner syndrome. The clinical context and clinical interview can generally guide the diagnosis which must be confirmed by clinical examination and complementary investigations (ENMG and/or MRI).

Although typical forms of nerve root pain do not raise any major diagnostic problems, some cases limited to distal paraesthesiae can be mistaken for TOS. The examination of the cervical spine, testing of deep tendon reflexes, Spurling manoeuvre and ENMG generally clarify the diagnosis. The possibility of referred pain from posterior joint or myofascial disorders of the anterior scalenus or of the sterno-cleido-mastoid muscles must also be considered. Finally, cervical spondylotic myelopathy can initially present with distal upper limb symptoms. The diagnosis can be corrected by a history of cervicobrachial neuralgia and symptoms below the level of the lesion.

Of the various tunnel syndromes, ulnar nerve compression at the elbow raises the most difficult problems of differential diagnosis. Although both diseases can cause paraesthesiae on the ulnar border of the hand, ulnar nerve compression is associated with more clearly systematized disorders (little finger and ulnar hemi-pulp of the ring finger). ENMG can eliminate this diagnosis. Carpal tunnel syndrome only raises diagnostic difficulties in atypical forms. Most importantly, carpal tunnel syndrome is so common that the diagnostic work-up should be continued in the presence of atypical symptoms, even when ENMG is suggestive of carpal tunnel syndrome. Note that Phalen's test can be positive in the presence of isolated TOS [26].

Rotator cuff syndrome can usually be easily distinguished from TOS on clinical examination. The diagnosis may be more difficult in the presence of degenerative disease in the same upper limb. In diffuse idiopathic pain syndrome or fibromyalgia, pain is both peripheral and axial, but some authors consider "disputable TOS" to be a particular form of fibromyalgia.

Finally, certain clinical forms or early stages of central nervous system diseases can be responsible for upper limb pain, paraesthesiae or distal muscle atrophy (syringomyelia, multiple sclerosis, amyotrophic lateral sclerosis). Cervical spondylotic myelopathy and certain peripheral neuropathies can present with atrophy of the intrinsic muscles of the hand, systematically requiring a complete neurological examination and ENMG.

All causes of upper limb pain can therefore be considered, but these causes can be simply concomitant diseases (tunnel syndrome or tendinitis) rather than true differential diagnoses. Recognition of this syndrome can avoid a number of unnecessary examinations or operations. However, evaluation of the psychosocial setting is essential, as these patients are known to frequently present an “unusual” psychological profile. Gockel showed that patients with TOS presented sympathetic hyperreactivity compared to a control population [38]. Socioeconomic or affective situations likely to lower pain tolerance or promote the emergence of “reactive” disorders must also be detected. As in all upper limb pain syndromes, stress, a certain individual susceptibility and poor motivation can be involved [39]. The individual perception of symptoms clearly differs as a function of the context in which they occur [32].

Treatment

Three types of treatment can be proposed: preventive measures, rehabilitation and surgery.

1/ Preventive measures are essential to correct or eliminate any risk factors identified, particularly in the workplace, as discussed below [33]. The use of orthoses has also provided useful results on distal symptoms in some patients [40].

2/ Rehabilitation was performed for many years according to Peet’s protocol [41], but a slightly modified protocol has often been used over recent years, comprising an initial analgesic and muscle relaxant phase that appears to give better results (especially in painful

neurological from). Correctly conducted rehabilitation can provide prolonged relief of symptoms in about 2/3 of patients [42, 43]. It is especially effective on proximal pain [30]. In refractory forms, **multidisciplinary management** is essential with evaluation of the various factors participating in maintenance of chronic pain and a retraining programme like those proposed in chronic low back pain [44].

3/ **Surgery** remains very controversial, even in relation to the various techniques [4]. Schematically, some authors are in favour of supraclavicular scalenectomy, while others are in favour of transaxillary resection of the first rib [6, 22, 37]. Scalenectomy appears to be associated with a lower success rate, and the results obtained also tend to wane with time [45]. It can be responsible for certain vascular and neurological complications and appears to be associated with a higher incidence of postoperative reflex sympathetic dystrophy. A cervical approach is especially indicated for resection of a superficial cervical rib, otherwise we consider transaxillary resection of the first rib to be the most appropriate method [6], as it is an effective procedure provided all nervous and vascular structures are completely released. However, this surgery is difficult and not devoid of certain risks with a high morbidity. A combination of the two techniques can sometimes be necessary [6].

Thoracic outlet syndrome and the workplace

Certain physical factors, especially dynamic factors, can predispose to TOS, particularly its painful forms. Many jobs involve repetitive movements and certain postural constraints such as jobs requiring use of the arms in elevation (hairdressers, switchboard operators, assembly lines, etc.). Repetitive movements with the upper limb raised, in antepulsion or abduction, carrying heavy loads on the shoulder or with the arm outstretched can induce compressions due to closure of the thoracic outlet and intermittent pressure on the brachial plexus (industrial workers, cashiers). Postural factors incriminated in sedentary jobs are mainly prolonged

cervical flexion (protraction of the head), and abduction and internal rotation of the shoulders (computer workers, musicians). These types of working position can induce an imbalance of muscles or the peripheral nervous system (tunnel syndromes). Use of the hand to move loads requires balanced control of all muscles of the upper limb and shoulder girdle. Many work-related overuse syndromes can therefore be associated with secondary TOS.

A review of the epidemiological literature, based on the **Pubmed** and **Embase** databases and using “thoracic outlet syndrome” and “occupational” as key words, limited to the human adult population and articles in French and English, looking for demonstrated occupational risk factors associated with thoracic outlet syndrome gave disappointing results. Fifty five articles were identified on the basis of these criteria. The second step consisted of reading the titles, abstracts, or articles according to their relevance in order to only include epidemiological studies (excluding case reports and editorials). Cross-references also identified several other articles. Ten articles were finally selected. Most of these articles concerned painful neurological forms of TOS with no neurological deficits, apart from the study by Sällström [18]. Diagnostic criteria, when they were defined, were very diverse, ranging from nonspecific tests (pain and limitation of neck and upper limb movements for Ohlsson [43], pain or paraesthesia of the upper limb for at least one week or once a month during the last 12 months for Battavi [47]), or a combination of symptoms and a clinical examination including specific stress tests [19, 48, 49].

The first result is the important prevalence in the workforce in these selected papers. In a sample including 191 workers (industry and service workers) with symptoms from the cervicobrachial region, 18% had TOS symptoms (27% of women and 11% of men) [18]. Pascarelli and Hsu found a 70% prevalence in a population workers with upper-limb

complaints of computer users and musicians (70% and 28 % respectively [19]), with a diagnosis including stress tests.

Postural constraints specific to certain occupations appeared to be possible risk factors according to these studies, especially the use of music instruments or video display screens (cashiers, secretarial work using a computer). Pascarelli and Hsu suggested that TOS related to postural constraints could predispose to other musculoskeletal disorders via a cascade compression mechanism (double crush syndrome) [19]. The role of this postural component in the pathogenesis of TOS must be considered not only in people working with their hands raised above the horizontal plane, such as painters, masons or forestry workers, but also people with jobs requiring retraction of the shoulders and rotation of the neck or working with the upper suspended, such as dentists, physiotherapists, hairdressers or musicians [50, 51]. Sällström and Schmidt [18] reported, in addition of a high prevalence of TOS, 2% of forms with severe symptoms. Prevalence rates according to job category could not be studied due to methodological limitations of the study. Hagberg and Wegman [49], in their review on musculoskeletal disorders of the shoulder, reported an excess risk of TOS in construction workers exposed to vibrations.

Overall, these studies presented many methodological limitations and no conclusion can be drawn concerning a significant association between TOS and occupational exposure (and therefore possible worker's compensation, except in particular cases).

The management of work-related TOS presents a number of specificities that should be known by physicians and occupational therapists. Key message is a multidisciplinary approach with medical care (as previously discussed) and workplace prevention. Preventive measures must be applied to correct or eliminate risk factors such as carrying heavy loads, correction of certain postures (arm in abduction-anteversion), regular rest breaks, etc. Global

management of workplace aspects is essential in view of the importance of mental health determinants involved in the pathogenesis of musculoskeletal disorders, mediation of pain processes and consequences of the pain in terms of work incapacity and disability [53]. Prognostic factors also depend on the social context of multidisciplinary management: a surgical series in the USA showed that the main factor preventing return to work was related more to psychosocial working conditions than to the operation itself [53]. Other associated overuse syndromes must also be managed after analysing the risk factors involved.

Conclusion

Apart from vascular and objective neurological deficit forms, TOS is a complex disease in terms of its aetiologies, pathophysiology, diagnosis and management. However, even functional, painful forms can be suspected by clinical examination and identification of risk factors. When in doubt, a multidisciplinary approach by specialized teams can allow early diagnosis and management of these patients including work rehabilitation taking into account prevalence in the workforce. Despite the low level of evidence, there is a possible link between the workplace and the individual in the pathogenesis and course of this disease. Further studies with clear definition, standardized exposure assessment should be conducted to study the role of occupational factor in the thoracic outlet syndrome.

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