Surgical treatment of thoracic outlet syndrome in young adults: single centre experience with minimum three-year follow-up

Pietro Ciampi · Celeste Scotti · Simonetta Gerevini ·
Francesco De Cobelli · Roberto Chiesa ·
Gianfranco Fraschini · Giuseppe M. Peretti

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Abstract Thoracic outlet syndrome is an often misdiagnosed syndrome which consists of a neurovascular compression at the upper thoracic outlet. The clinical presentation can be variable, ranging from mild symptoms to venous thrombosis and muscle atrophy. Many aetiologies, both congenital and acquired, related either to bony or soft tissue anomalies, have been associated with this syndrome. As a consequence, the diagnosis is often challenging and sometimes it can be obtained only with surgical exploration. Additionally, no

specific clinical test is considered diagnostic of thoracic outlet syndrome. However, the recent advances in imaging techniques together with a careful clinical evaluation give the surgeon the chance to recognize the constricting anatomy before surgery in many cases. No standard surgical procedure has been identified; however, in literature the largest series have been treated with transaxillary first rib resection. Here we report our experience in the surgical treatment of this syndrome with a minimum follow-up of three years. Our approach consists of performing a supraclavicular decompression without routine first rib resection. This allows for identifying and removing the constricting anatomy in most cases, with satisfactory results in 96.9% of patients and a low complication rate.

P. Ciampi · C. Scotti · G. Fraschini (✉) · G. M. Peretti (✉)
Department of Orthopaedics and Traumatology,
San Raffaele Scientific Institute,
Via Olgettina 60,
20132, Milan, Italy
e-mail: fraschini.gianfranco@hsr.it

G. M. Peretti
e-mail: peretti.giuseppe@hsr.it

S. Gerevini
Department of Neuroradiology, San Raffaele Scientific Institute,
Via Olgettina 60,
20132, Milan, Italy

F. De Cobelli
Department of Radiology, San Raffaele Scientific Institute,
Via Olgettina 60,
20132, Milan, Italy

R. Chiesa
Department of Vascular Surgery, San Raffaele Scientific Institute,
Via Olgettina 60,
20132, Milan, Italy

G. M. Peretti
Faculty of Exercise Sciences, University of Milan,
Via Kramer 4/A,
20129, Milan, Italy

Introduction

The term “thoracic outlet syndrome” (TOS) was introduced by Peet et al. in 1956 [15]. TOS consists of a myriad of symptoms depending on compression of the neurovascular bundle of the upper limb as it passes through the thoracic outlet [11, 18, 22]. As a matter of fact, TOS represents a spectrum of disorders encompassing at least four related syndromes: (i) neurogenic TOS in cases of compression of the brachial plexus, (ii) arterial TOS in cases of compression of the subclavian artery, (iii) venous TOS in cases of compression of the subclavian vein, and (iv) disputed TOS, a non-specific type of TOS with unclear origin. Many aetiologies, both congenital and acquired, may determine a compression to the neurovascular bundle: bony anomalies such as cervical ribs, transverse mega-apophyses, first rib abnormalities, clavicle non-unions; soft tissue anomalies such as cervical muscles hypertrophy or fibrous bands;

posture problems such as drooping or sagging of the shoulders, or even poor posture due to large breast [18]. In some cases (disputed TOS) no obvious causes are identified [30]. Neurogenic symptoms are the most common and they account approximately for 95% of all TOS [4, 24]; interestingly, in the US series this condition appears to be more represented than in the European ones [2, 11]. As a matter of fact, the true incidence of TOS is unknown; however, it has been estimated as 5:100,000 per year in the United Kingdom [26].

The lack of a diagnostic test for TOS makes the management of this syndrome, either surgical or non-operative, very challenging; consequently, the treatment of TOS is debated and long-term results are poorly documented [12, 29, 30]. In particular, in disputed TOS the diagnostic and therapeutic processes are even more challenging, as no evident compression to the neurovascular bundle can be identified. Many surgical approaches to the thoracic outlet have been described: transaxillar [20], supraclavicular [8, 27], infraclavicular [7], and posterior [9]. Although the largest series reported advocate transaxillary first rib resection as the standard procedure for TOS, with more than 90% success rate [21, 28], comparable results have been reported without routine first rib resection [11, 23].

The aim of this retrospective study was to outline the management of this complex syndrome, focusing on the diagnostic process, on the corresponding surgical findings, and on the results of surgical decompression of disputed and undisputed TOS cases.

Materials and methods

A series of 50 patients treated for TOS by the same surgeon (GF) in our department from 1999 to 2006 is analyzed here. In this period, more than 500 patients with symptoms such as pins and needles in the arm, severe pain, loss of function, muscle wasting or arm claudication were evaluated by the Shoulder Unit of our department for the diagnosis of TOS. Inclusion criteria for undisputed TOS were presence of suggestive symptoms and imaging evidence of compression to the neurovascular bundle. Persistence of suggestive symptoms after a six-month physical therapy program, without any evidence of compressive structure, was considered as inclusion criteria for disputed TOS. Accordingly, patients were divided in two groups: a non-disputed TOS group (30 cases) and a disputed TOS group (20 cases).

Forty-eight patients (38 women and ten men) with a mean age of 29 years (range 20–47) were included in this study and underwent surgical treatment for TOS. Two patients had bilateral symptoms. Symptoms involved the dominant hand in 48 cases (96%) and the non-dominant hand in two (4%). The mean duration of symptoms was

38 months (range 4–240). Patients were evaluated by careful analysis of duration and entity of signs and symptoms, either vascular or neurologic, and by compression and provocative tests [17]. We used the Tinel's sign and a panel of four provocative tests (Fig. 1), namely, Adson's test (modified by Rayan) [18], Roos' test, the costoclavicular manoeuvre, and the hyperabduction manoeuvre. Great care was taken to duplicate the symptoms with these tests and to emphasize the nature of the symptoms, either vascular or neurologic.

Regarding diagnostic techniques, all patients underwent plain X-ray of the cervical spine in order to identify bony anomalies and magnetic resonance (MR) of the neck and of the soft tissues of the thoracic outlet in order to rule out ruptured disk, spinal stenosis, or other neurologic abnormalities. MR was performed dynamically, in neutral position and in hyperabduction, thus duplicating the position which usually reproduces the symptoms, with neurographic (STIR) and angiographic sequences (T1 3D Gradient Echo), to carefully evaluate the scalene triangle and the costo-clavicular space. Electromyography (EMG) was carried out in all patients with the only purpose of facilitating the differential diagnosis process, as this exam is usually negative in patients with TOS.

The surgical treatment consisted of a supraclavicular decompression in all patients with an infraclavicular exploration in five cases (10%) and a step-cut osteotomy of the clavicle in one case (2%). Outcome was evaluated by the questionnaire developed by Bhattacharya et al., which consists of a four-degree (excellent, good, fair, poor) scale [1]. The procedure was considered successful in cases of excellent, good or fair results [1, 23]. The questionnaires were obtained during routine follow-up visits in the outpatient department; otherwise patients were contacted by telephone by a member of the Shoulder Unit. Follow-up ranged from three years to six years (mean 4.5 years).

Results

Clinical findings

Symptoms were mostly mixed, both vascular and neurologic. However, in the non-disputed group, presentation was predominantly arterial (90% of patients) with positive provocative tests, arm claudication and coldness. These symptoms were exacerbated during overhead activities. In the disputed TOS group, presentation was predominantly neurological (75% of patients) with low-plexus plexopathy or diffuse symptoms, e.g. hypoesthesia with ulnar or whole hand distribution; pain in the supraclavicular fossa, posterior neck and medial arm and forearm; fatigue and weakness of the intrinsic muscles of the hand and difficulty with fine motor tasks.

Fig. 1 The four basic clinical tests we included in our routine evaluation of patients with thoracic outlet syndrome (TOS). For each of them, a positive test is indicated by absence of the radial pulse or by the presence of any neurologic sign. **a** Adson's test. The examiner locates the radial pulse. The patient extends the neck while the examiner extends the arm and keeps it extra-rotated. **b** Hyperabduction test. The patient is asked to keep the arm hyperabducted, the shoulder extra-rotated and the elbow extended. **c** Costoclavicular manoeuvre. The examiner locates the radial pulse and draws the patient's shoulder down with elbow extended, as the patient lifts their chest in an "at attention" or "military" posture. **d** Roos' test. The examiner locates the radial pulse, elevates/extra-rotates the shoulder while flexing the elbow

Diagnostic studies

Plain X-rays were positive in all patients with bony anomalies. Dynamic MR identified the constricting anatomy in all patients of the non-disputed group. On the other hand, EMG was always negative in patients of both groups. In patients of the non-disputed group the diagnosis was always performed with imaging techniques: 18 patients (36%) were affected by cervical rib (Fig. 2), three patients (6%) by abnormal first rib, four patients (8%) by supraclavicular lipoma (Fig. 3), three patients (6%) by transverse megapophysis, and two patients (4%) by clavicle non-union.

Surgical findings

In patients of the non-disputed group, pre-operative imaging assessment always corresponded to the surgical findings. In patients of the disputed group, the diagnosis was performed during surgical exploration: 15 patients (30%) were affected by anterior scalene muscle anomalies, 11 of which presented hypertrophic muscles (Fig. 4), and four a falciform insertion (Fig. 5); one patient (2%) was affected by anomalous band (type 3 according to Roos'

classification) [20]; and four patients (8%) were affected by perineural fibrosis. All these four patients had a history of cervical trauma. Three patients (6%) of the disputed group had a post-stenosis pseudoaneurysm of the subclavian artery; one of them was replaced with a vascular prosthesis by the vascular surgeon. Surgical exploration was performed by a supra-clavicular approach in 45 patients (90%) (Figs. 2, 3, 5), and by a combined supra- and infra-clavicular approach in five patients (10%) (Fig. 4).

Clinical outcome

The outcome questionnaire was obtained by 48 patients (96%) as two were lost to follow-up and could not be contacted. Table 1 shows the results of this evaluation after a three-year minimum follow-up. Using the definition of success as excellent + good + fair [1, 23], the overall success rate was 95.9% (96.4% for the undisputed group and 95% for the disputed group); with a definition of success as excellent + good the success rate was 79.2% (82.1% for the undisputed group and 75% for the disputed group). One patient (2%) had revision surgery due to early recurrence of symptoms such that during the second

Fig. 2 Undisputed thoracic outlet syndrome (TOS) due to cervical rib. **a** Pre-operative X-ray demonstrating the cervical rib (arrow). **b** Pre-operative dynamic angiography showing a partial reduction of the arterial flow during hyperabduction (arrow). **c** Intra-operative expo-

sure showing the vicinity of the cervical rib to the subclavian artery. **d** Isolation and protection of the subclavian artery and brachial plexus in order to perform the osteotomy and resection of the cervical rib

surgery adhesences around the brachial plexus were identified and released.

The mean hospital stay was four days (range 3–6 days) and there were three postoperative complications (6.2%). One patient (2%) developed a mild recurrence of symptoms but did not undergo a second surgical procedure; one patient (2%) who underwent first rib resection developed a small apical pneumothorax but no chest drainage was applied; and one patient (2%) developed a neuroproaxia which resolved completely after one month. Neither major complications nor wound infections were observed.

Discussion

TOS is an often misdiagnosed cause of arm and neck pain and disability depending on compression to the neurovascular bundle of the thoracic outlet [13]. The complexity in this diagnosis is due to the myriad of symptoms and the different aetiologies which characterize this syndrome and, on the other hand, in the lack of a specific diagnostic test or exam [11]. The differential diagnosis includes cervical radiculopathy, supraclavicular fossa pathology, trauma, tumors (especially pulmonary apical masses), brachial

Fig. 3 Undisputed thoracic outlet syndrome (TOS) due to lipoma occurring in the supraclavicular fossa. **a** Pre-operative MR demonstrating the lipoma (arrow). **b** Intra-operative exposure showing the

lipoma. **c** Intra-operative exposure after lipoma excision showing the normal anatomy of the thoracic outlet. In particular, no anomalies of the anterior scalene muscle are evident

Fig. 4 Disputed thoracic outlet syndrome (TOS) due to anterior scalene muscle anomaly. **a** Pre-operative dynamic angiography showing a complete stop of the arterial flow behind the clavicle during hyperabduction (*arrow*). **b** Intra-operative exposure showing scalene muscle hypertrophy (*arrow*). **c** Intra-operative exposure

showing the step-cut osteotomy of the clavicle performed in order to achieve a better view of the subclavian artery and allow for the subsequent vascular reconstruction. **d** Subclavian artery pseudoaneurysm (*arrow*). **e** Vascular reconstruction with a synthetic vascular graft

neuritis and complex regional pain syndrome type I (reflex sympathetic dystrophy) [14]. Moreover, associated double-crush syndromes, presenting a second compression distally to that involving the nerve at the thoracic outlet, may account for 44%, thus making the diagnosis even more challenging [31]. As a matter of fact, the diagnosis is usually performed clinically, while imaging and electrodiagnostic studies are useful tools in the differential diagnosis process and in identifying the cause of the compression in some cases [18].

As far as the electrodiagnostic studies are concerned, EMG was negative in all patients in our series. This was because symptoms such as chronic denervation of tenar and hypotenar muscles are usually present only in the most severe forms of TOS, characterized by a long history of

weakness and atrophy. However, EMG may be helpful in excluding other proximal or distal nerve compressions. On the other hand, MR allows performing a complete evaluation of the muscular, vascular and nervous components of the thoracic outlet and should be considered as the method of choice for the evaluation of a patient with TOS [3, 5, 6]. The study should be performed in neutral position and in hyperabduction in order to highlight the compression [3, 5, 6]. Additionally, it utilizes no ionizing radiation and gadolinium as contrast material which has fewer side effects than iodinated ones used for digital angiography. Recently, we included multidetector-CT among the preoperative diagnostic tools [10]. However, this technique presents some limitations in the evaluation of patients with TOS, for example, it can not carry out a fine analysis of the

Fig. 5 Disputed thoracic outlet syndrome (TOS) due to anterior scalene muscle anomaly. **a** Normal pre-operative X-ray. **b** Dynamic Angio MR showing a stop in the arterial flow during hyperabduction (arrow). **c** Sagittal MR study of the costoclavicular space with STIR sequence showing reduction of the space between the clavicle and the

first rib during hyperabduction (arrow). **d** Intra-operative exposure showing the falciform insertion of the anterior scalene muscle. **e** Intra-operative exposure after scalenectomy demonstrating the release of the subclavian artery

brachial plexus due to the limited CT contrast resolution and it is an ionizing radiation technique [6]. Consequently, we perform multidetector-CT only in cases of bony anomalies and vascular symptoms, always in combination with MR.

Regarding the treatment of TOS, both nonoperative and surgical approaches have been described [18]. Physical therapy consists mainly of shoulder girdle elevator muscle strengthening and pectoral muscle stretching [12]. Nevertheless, nonoperative management is often effective in

Table 1 Summary of surgical procedures performed in our series and clinical outcome

Procedure	Number performed	Excellent/good	Fair	Poor
Overall (undisputed TOS)	28	23 (82.1%)	4 (14.3%)	1 (3.6%)
Cervical rib resection	16	13 (81.3%)	2 (12.5%)	1 (6.2%)
First rib resection	3	3 (100%)		
Lipoma excision	4	4 (100%)		
Mega-apophysis resection	3	2 (66.6%)	1 (33.3%)	
Clavicle nonunion treatment	2	1 (50%)	1 (50%)	
Overall (disputed TOS)	20	15 (75%)	4 (20%)	1 (5%)
Scalenectomy	15	12 (80%)	2 (13.4%)	1 (6.6%)
Excision of band	1		1 (100%)	
Brachial plexus release	4	3 (75%)	1 (100%)	
Total (disputed and undisputed TOS)	48	38 (79.2%)	8 (16.7%)	

TOS thoracic outlet syndrome

patients with posture problems, but it is ineffective in cases with structural anomalies as it can, at best, delay surgery [18]. In our experience three patients (6%) who were admitted to surgery after several months of ineffective physical therapy presented at surgical exploration a subclavian artery psuedoaneurysm and one of them required vascular replacement with prosthesis by the vascular surgeon. Consequently, we do not prolong physical therapy anymore, if ineffective after 12 weeks [18].

As far as the surgical management is concerned, no standard surgical approach has been identified yet [11]. However, the largest series published in literature report a high success rate with transaxillary first rib resection [21, 28]. The rationale of this approach is that the first rib forms the common denominator for all causes of nerve and artery compression in this region, so that its removal generally improves symptoms [20]. On the other hand, some serious complications, such as winging and snapping of the scapula and pneumothorax, have been associated with this procedure [2, 21, 23, 25, 31], and a high rate of recurrence has been reported by some studies [16, 23]. Consequently, in order to reduce the rate of complications and recurrence, the supraclavicular approach has been advocated to perform first rib resection and scalenectomy [7, 23, 25].

As a matter of fact, the supraclavicular approach demonstrated to be a safe and effective procedure, characterized by a shorter operative time and having a complication rate lower or comparable to that of transaxillary first rib resection [15, 23, 25]. Moreover, the transaxillary approach does not permit performing vascular repair in cases of subclavian artery pseudoaneurysm. On the other hand, the supraclavicular approach allows for a wide view of the subclavian artery when performing also the infraclavicular exploration combined with a clavicle step-cut osteotomy. In addition, the supraclavicular approach allows performing intraoperative provocative manoeuvres before and after the decompression, in order to identify the site of compression and to verify the effectiveness of the release. This is particularly useful in the disputed cases in which no aetiology is identified with the imaging studies; we recognized a constricting anatomy in all patients with disputed TOS undergoing this procedure. In our series, we registered an overall success rate of 95.9% and an 8% (4 cases) overall rate of complication with one (2%) early recurrence, treated successfully with a second extended procedure, and three (6%) minor complications. These results are comparable to those reported in literature for supraclavicular or transaxillary first rib resection [8, 15, 22] and to those previously published for supraclavicular decompression [11, 19, 23, 25].

Our results demonstrate that satisfactory results can be achieved with supraclavicular decompression, also in the disputed cases, without routine first rib resection. This procedure allows for performing exploration of the supra-

clavicular region, with accurate identification and release of any constricting structure, and combined vascular repair if needed. In addition, with this approach the first rib can be resected if recognised as the cause of the compression. However, these results have to be confirmed at long-term follow-up and by a larger series with a randomized multicenter comparative study, as most published studies regarding this approach account for a small number of patients and utilize different outcome criteria [1, 11].

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