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Original article

Long-term functional outcomes after surgical treatment of nonspecific thoracic outlet syndrome: Retrospective study of 70 cases at a mean of 8 years' follow-up

Résultats cliniques à long terme après chirurgie du syndrome du défilé cervico-thoraco-brachial à expression neurologique subjective : étude rétrospective de 70 cas au recul moyen de 8 ans

A. Perchoc [a,*], C. Andro [a], H. Letissier [b], P. Schiele [c], D. Le Nen [b]

[a] *Hôpital d'instruction des armées Clermont-Tonnerre, rue Colonel-Fonferrier, 29240 Brest Armées, France*
[b] *Centre hospitalier régional universitaire de la Cavale Blanche, boulevard Tanguy-Prigent, 29200 Brest, France*
[c] *Hôpital d'instruction des armées Desgenettes, 108, boulevard Pinel, 69003 Lyon, France*

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ABSTRACT

Our objective was to study the clinical and functional outcomes after surgical treatment of nonspecific thoracic outlet syndrome (TOS) using a supraclavicular approach. We included every patient with TOS who was treated surgically by the same surgeon at a single hospital between 1999 and 2014 with a minimum follow-up of 4 years. The primary outcome was the overall evaluation of the function by the QuickDASH Score. Secondary outcomes included functional, subjective outcomes, pain levels, and neuropathic features. The clinical outcome of 70 cases was evaluated with a mean follow-up of 8.5 years. The improvement in the QuickDASH Score was significant with an average change of 38 points. Mean pain intensity was reduced postoperatively by 1.6 points from 5.1/10 to 3.5/10. Persistent pain rate at 4/10 or more remained in 56% of cases. These cases were mostly neuropathic. The surgical procedure significantly reduced the number of patients suffering from paresthesia, numbing or weakness. The possibility of sequelae and the persistence of neuropathic pain means the surgical indication should be limited to patients with significant functional disability despite well-conducted rehabilitation. The long-term functional outcomes in patients undergoing TOS was mostly good but could be improved by addressing residual neuropathic pain symptoms.

RÉSUMÉ

Notre objectif était d'étudier les résultats cliniques et fonctionnels à long terme des syndromes neurologiques douloureux du défilé cervico-thoraco-brachial (DCTB) opérés par voie supraclaviculaire. Les patients étudiés avaient été opérés entre 1999 et 2014 d'un syndrome neurologique non déficitaire du DCTB par le même opérateur dans un seul centre avec un recul d'au moins quatre ans. Le critère de jugement principal était l'évaluation globale de la fonction par le Score QuickDASH. Les critères de jugement secondaire comprenaient les résultats fonctionnels, subjectifs, l'analyse des douleurs et des signes neuropathiques. Le résultat clinique de 70 interventions a été évalué avec un recul moyen de 8,5 ans. L'amélioration du Score QuickDASH a été significative avec une réduction moyenne du score de 38 points. L'intensité moyenne de la douleur a été réduite en postopératoire de 1,6 point passant de 5,1/10 à 3,5/10. Un fond douloureux supérieur à 4/10 restait présent dans 56% des cas. Ces douleurs étaient

* Auteur correspondant: Hôpital d'instruction des armées Clermont-Tonnerre, rue Colonel-Fonferrier, 29240 Brest Armées, France
E-mail address: perchocarnaud@gmail.com (A. Perchoc).

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majoritairement de type neuropathique. Il était trouvé une amélioration clinique significative post-opératoire associant réduction des paresthésies, de l'engourdissement, du manque de force et des douleurs. Les possibilités de séquelles et de persistance de signes neurologiques impliquent de réserver l'indication opératoire aux malades présentant une importante invalidité fonctionnelle malgré une rééducation bien menée. Le devenir fonctionnel sur le long terme des patients opérés d'un syndrome du DCTB est en majorité satisfaisant, mais une prise en charge des douleurs neuropathiques résiduelles pourrait l'améliorer.

1. Introduction

Thoracic outlet syndrome (TOS) is a condition related to compression of neurovascular bundles in the upper limb. While the vascular and true neurogenic forms have an accepted diagnostic and therapeutic pathway [1], the subjective (nonspecific) forms are more controversial. This is due to the varied clinical presentation, large spectrum of lesions and variety of compressive causes [2]. Here, we report on our experience with the diagnosis and surgical treatment of 70 cases of nonspecific TOS.

The aim of this study was to describe the long-term functional outcomes and relationships with an underlying compressive cause and to look for prognostic factors. The working hypothesis was that surgical treatment of nonspecific forms of TOS through a supraclavicular approach will reduce pain and improve long-term function. The primary endpoint was the QuickDASH as a measure of functional outcome.

2. Patients and methods

2.1. Patients

This was a single-center, retrospective study. We included patients operated for nonspecific TOS through a supraclavicular approach who had at least 4 years' follow-up.

The diagnosis was made based on the consistency of the disease history, symptoms and clinical examination; this information was used to rule out differential diagnoses such as myofascial pain syndrome [3]. The symptoms developed gradually in most cases with the appearance of paresthesia and numbness of the upper limb and/or ulnar side of the hand combined with "dead arm syndrome". The Roos test, positional nature of symptoms, pain elicited by palpation of the scalene muscles and Tinel sign were determined.

The surgical indication was made after conservative treatment had failed [4,5] and in the presence of disabling pain that significantly impacted the patient's function. In cases of double crush syndrome [6] based on clinical presentation and electromyography data [7], peripheral surgical procedure was done first. The TOS release procedure was delayed by at least 6 months.

All patients had a preoperative work-up:

- X-rays of the cervical spine to look for bone abnormality;
- X-rays of the thorax area to rule out a tumor;
- electromyography to look for a peripheral outlet condition or involvement of the suprascapular nerve;
- Doppler ultrasonography with dynamic movements.

If the latter was abnormal, we requested a CT angiography with three-dimensional reconstruction. If a regional tumor was suspected, we ordered an MRI of the supra- and infraclavicular region. If a rotator cuff ailment was suspected, ultrasonography of the shoulder was ordered.

All patients were operated by the same experienced surgeon. The procedure was done by the supraclavicular approach consisting of anterior and midline scalenectomy with release and evaluation of the subclavian artery, and then release of the brachial plexus. Any bone elements causing compression, such as a cervical rib or malformed C7 process, or compressive muscular or ligament elements were resected. If there was no obvious intraoperative abnormality, the first rib was resected.

2.2. Data collection

Functional outcomes were collected preoperatively and then at the last follow-up using a questionnaire incorporating the QuickDASH Score [8]. The patient's postoperative satisfaction was labeled as very satisfied, satisfied, not satisfied or disappointed. We looked for prognostic factors. Return to work was determined. Pain was evaluated on a simple numeric scale. We looked for postural triggering. The neuropathic nature of the pain was detected postoperatively with the DN4 Questionnaire [9]. We recorded the presence of subjective neurological signs such as the presence of paresthesia, numbing, weakness, dead arm syndrome and information about serious postoperative complications.

2.3. Statistical analysis

Student's *t*-test was used to analyze quantitative variables and Fisher's exact test was used to analyze qualitative variables. For easier readability, decimals were removed from the results expressed in percentages. Rstat® software was used for the statistical analysis.

3. Results

We identified 79 cases performed between 1999 and 2014. The study flow chart is shown in Fig. 1. Nine patients were lost to follow-up, leaving 70 cases for analysis. The mean follow-up was 102 months (95% CI: 48–156), or 8.5 years. The patients' demographics and clinical data are summarized in Table 1. The anatomical abnormalities discovered intraoperatively are listed in Table 2.

The patients operated for TOS had positional triggering of symptoms preoperatively in 91% of cases. They had a positive Roos sign in 97% of cases, while 87% had pain upon palpation of the scalene muscles and 81% had a positive Tinel sign. The mean QuickDASH was 89 preoperatively and 51 after the procedure. The 38-point mean decrease was statistically significant ($P < 0.05$). We found that 24% were very satisfied with the outcome, 53% were satisfied, 17% were somewhat satisfied and 6% were disappointed. Fifty-seven patients (81%) said they would undergo the procedure again.

There was no statistical correlation between the outcome and sex, work-related trigger, psychological predisposition, return to

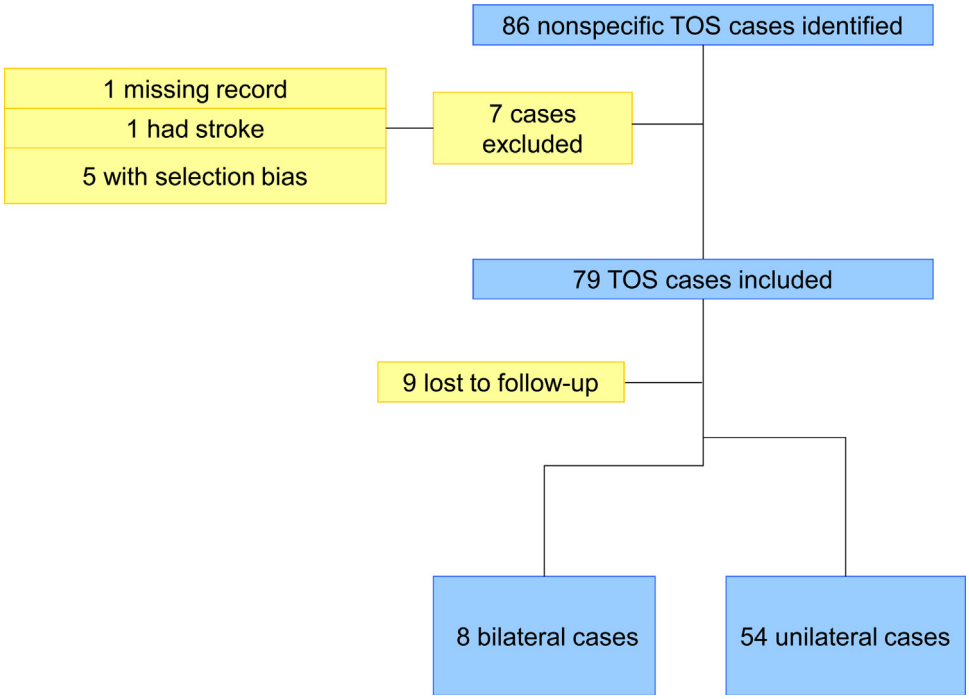

Fig. 1. Flow chart summarizing the inclusions for this study.

Table 1
Demographics and clinical features of the study population.

Variable	Value
Sex, No. of patients	32 (46%)
Male	38 (54%)
Female	
Handedness, No. of patients	
Left-handed	7 (10%)
Right-handed	60 (86%)
Ambidextrous	3 (4%)
Dominant side involved	37 (53%)
Mean age when symptoms appeared, years ± standard deviation	37 ± 9.9
History of depression or anxiety, No. of patients	
Chronic anxiety	6 (9%)
Depression	17 (24%)
Antidepressant therapy	11 (16%)
Suicide attempt	3 (4%)
Post-traumatic stress syndrome	1 (1%)
Intensity of preoperative pain, No. of patients	
Severe	32 (49%)
Significant	22 (33%)
Moderate	7 (11%)
Slight	5 (7%)
Appearance of symptoms, No. of patients	
Progressive	53 (77%)
Sudden	7 (10%)
Step changes	8 (13%)
Triggering factor, No. of patients	
Trauma	13 (18%)
Occupation	45 (65%)
Sports activity	3 (4%)
Unusual effort	2 (3%)
No obvious factor	7 (10%)
Bilateral involvement	8 (11%)
Distribution of pain, No. of patients (%)	
Diffuse	29 (41%)
Localized to hand	24 (34%)
Localized to shoulder	17 (24%)
Also present at night	35 (70%)

Table 2
Intraoperative abnormalities observed in the 70 cases.

Abnormality	Number (%)
Dynamic narrowing	30 (43%)
Middle scalene hypertrophy	29 (41%)
Isolated	15 (21%)
Associated with another compressive disorder	14 (20%)
Cervical rib	1
Fibrous tract at middle scalene forming anterior fibrous rib	7
Agenesis of anterior scalene	1
Hypertrophy of anterior scalene	1
Costocostal ligament	1
Fibrous tract surrounding T1	1
Fibrous tract surrounding subclavian artery	1
Presence of cervical rib	13 (19%)
Isolated	7
Associated with another abnormality	6
Fibrous track joining cervical rib with 1st rib	5
Hypertrophy of middle scalene	1
Enlarged C7 process	1
Enlarged C6 and C7 process combined with false transverse rib	1
Other	
Costocostal and vertebrocostal ligament suppressing T1	11 (16%)
Agenesis of anterior scalene	1
Agenesis of plexus roots replaced by multitude of smaller nerves	1
Pre-arterial tubercle of the first rib bracketing the artery	1
Malunion of clavicle	1
Fibrous band of middle scalene compressing C8 and T1	1
Artery compression at anterior scalene by its bone attachment on either side	1
Transverse costal ligament	1
Plexus held in the superior interpedicle formations	1
Plexus is horizontal	1
Plexus has fibrosis	1
Posterior synostosis between 1st and 2nd rib	1

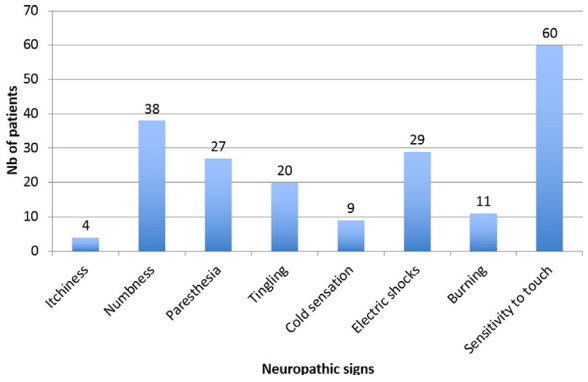

Fig. 2. Prevalence of neuropathic pain symptoms.

work, resection of the cervical rib or first rib and hypertrophic middle scalene.

Return to work was achieved in 63% of cases (22 in their prior occupation and 14 with adaptations, including 8 job changes). Eighteen patients were on disability, while five were unemployed and three retired.

The mean intensity of the preoperative pain was 5.1/10 (95% CI: 3–7) and the postoperative pain was 3.5/10 (95% CI: 1–6). The pain was completely gone in 29% of cases and was reduced in 51%. The pain was unchanged or worse in 20% of patients. Residual pain was often position-related: a specific position (abduction at more than 90°) triggered pain in 46 cases (66%). Twenty-nine patients (41%) had neuropathic pain, as they had a score of 4 or more on the DN4. The prevalence of various neuropathic pain signs is shown in Fig. 2.

The surgical procedure significantly reduced the number of patients suffering from paresthesia, numbing or weakness. There was fewer cases of dead arm syndrome after the procedure, but this change was not significant. These findings are described in further detail in Fig. 3.

As for complications, there were two cases of apical pneumothorax that required monitored only and two cases of moderate hemothorax. There were two cases of diaphragmatic paralysis due to phrenic nerve injury (3%) that resolved in a few weeks for one patient and in 12 months for the second patient. On the neurological side, injury to the supraclavicular branches of the superficial cervical plexus was responsible for scar-related or prethoracic hypoanesthesia or anesthesia in 22 cases (31%) or a painful area in the operated region in 7 cases (10%). One patient (1.4%) had an infection that required revision for sample collection and lavage, followed by antibiotic therapy. There were three cases of secondary adhesive capsulitis (4%); the condition resolved in two patients and persisted in the other patient. There were no vascular complications.

4. Discussion

Our study is significant because 70 cases were followed for an average of 8.5 years (range 4 to 15) and only 11% of cases were lost to follow-up. The sex ratio (54%) showed a slight predominance of females cases, which is typical for TOS [10].

The diagnosis of TOS is primarily clinical despite improvements in imaging modalities [10,11–13]. We used the same diagnostic criteria as in other published studies [14], in particular the Roos test, which has an 84% sensitivity [15]. Electroneuromyography is required to identify double crush syndrome [15,16]. We identified this condition during history taking in more than one out of two patients.

The triggering factor was often related to work (65%) and more rarely post-traumatic (18%). The non-dominant side was affected more often (53%), which is also consistent with published data [17].

The social and professional repercussions of TOS were significant: one-third of patients were disabled or unemployed, one-third had a career change, and only one-third were able to resume their prior job, often with adaptation. These outcomes are not as good as those described by Green et al. [18]. A change in occupation is often required because of disease progression. It is sometimes done to avoid the development or aggravation of TOS on the opposite side. This reorientation may be a bias in the progression of the symptoms because the cause is removed.

In other TOS studies, the outcome is typically evaluated by the surgeon. In our study, the functional outcome score (QuickDASH) was determined preoperatively and postoperatively. The QuickDASH, which was our primary endpoint, significantly improved after the surgery in 81% of cases (57 patients). The self-reported nature of this questionnaire removes the evaluation bias [19]. Given that other studies did not use a functional outcome score, we could only compare the rate of failure or poor results, i.e. patients who did not improve after the procedure. The findings from various studies on this topic are given in Table 3 [19,22–24,26,27,35–41]. The failure rate in our study was comparable to that of other published studies (20%).

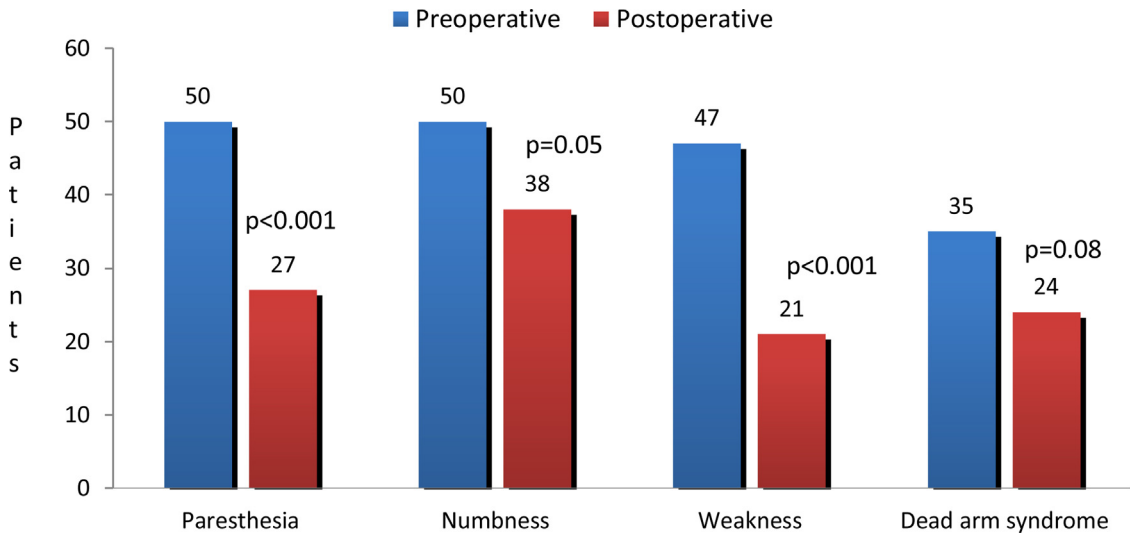

Fig. 3. Changes in functional signs following surgical treatment.

Table 3
Summary of TOS functional outcomes in the literature (K1: resection of first rib).

Study: author, year	Number of cases	Average follow-up	Procedure used Surgical approach and resection	% successful
Hempel et al., 1981 [26]	433		*Supraclavicular approach* K1 and scalenotomy	84.5%
Roos, 1982 [35]	93		*Supraclavicular ± Roos* Scalenectomy ± K1	79% in revision cases
Qvarfordt et al., 1984 [23]	191		*Roos approach* K1 + anterior scalenotomy	79%
			Supraclavicular approach Prior patients revised by TOS release	80% excellent, 20% good
			Combined approach: Roos and supraclavicular K1	95% excellent, 4% good
Narakas et al., 1986 [36]	75		*Roos approach* K1	82.5%
			Cervical approach K1	50%
			Cervical approach Cervical rib resection	77%
			Cervical approach Scalenotomy only	44%
Cormier and Kiefer, 1989 [22]	131	62 months	*Supraclavicular approach* Anterior scalenectomy, middle scalenotomy, K1	81%
Sanders and Pearce, 1989 [27]	668	60 to 120 months	*Roos approach* K1	69%
			Supraclavicular approach Anterior and middle scalenectomy	69%
			Supraclavicular approach K1 and scalenectomy	72%
Gockel et al., 1994 [19]	107	49 months	Scalenotomy + cervical rib resection ± K1 if revision	63%
Lindgren and Oksala, 1995 [37]	45	96 months	*Roos approach* K1 or cervical rib resection	43% healing rate
Goff et al., 1998 [38]	54	24 months	*Roos or supraclavicular* K1	48% good, 39% acceptable results
Urschel and Razzuk, 1998 [39]	2210, of which 222 were bilateral		*Roos approach* K1 + anterior scalenectomy	- 96% for C5-C6 involvement - 95% for mixed involvement - 95% for C8-T1 involvement
Athanassiadi et al., 2001 [40]	23		*Roos approach* K1	82%
Sanders and Hammond, 2002 [24]	42, of which 12 were bilateral		*Supraclavicular approach (most cases)* resection cervical rib ± scalenectomy ± K1	72%
Merle et al., 2011 [41]	368	58 months	*Supra- and infraclavicular (Cormier)* Anterior and middle scalenectomy, K1	88%
			Cervical and deltopectoral approach As above, without anterior scalenectomy and resection of coracoclavicular ligament	95%
Our study	70	102 months	*Supraclavicular approach* Anterior and middle scalenectomy ± K1 ± cervical rib resection	81%

We felt it was relevant to evaluate the effectiveness of the procedure by analyzing pain levels before and after the surgery. The mean intensity of preoperative pain was 5.1/10 (95% CI: 3–7) and postoperative pain was 3.5/10 (95% CI: 1–6). This corresponds to severe pain becoming moderate pain. In the study by Merle et al., 37% of patients had complete pain relief versus 29% in our study; pain was reduced in 50% of cases versus 51% in our study; it was unchanged in 13% of patients versus 20% in our study.

We found that 29 patients (41%) had neuropathic pain according to the DN4. There was a significant reduction in neurological symptoms postoperatively.

We recommend the DN4 questionnaire [9] be used preoperatively to detect and subsequently treat neuropathic pain.

We did not identify any prognostic factors. Thus, sex, work-related trigger, return to work, and psychological predisposition had no significant effect on the outcome. Contrary to our study, there are published reports of a statistical correlation between a poor outcome and major depressive episode [20]. This may be due to a lack of statistical power or excessive variability in our study population. It has been recommended that patients with a depressive condition not be operated on until psychiatric care has been successful [21].

In terms of anatomy, hypertrophy of the middle scalene was a common intraoperative observation (41% of cases), and had a higher rate than in the Cormier and Kieffer study (27%) [22]. A cervical rib was present in 19% of cases; this varies greatly in other studies from less than 1% [23] to up to 92% [24]. It is important to remember that these anatomical variations are common in the general population (95%) and often asymptomatic [25]. This is a risk factor predisposing the appearance of TOS, which may develop secondarily following neck trauma or due to a work-related trigger.

Using the supraclavicular surgical approach allowed us to effectively treat compressive disorders: anterior and middle scalene, neurolysis of brachial plexus, resection of cervical rib and/or first rib. [23] This approach tends to be preferable as it allows neurogenic TOS to be treated with less morbidity than the Roos approach [26] with fewer serious complications [27]. The outcome is not affected by the type of surgical approach and whether or not the first rib is resected. Our findings are consistent with those of Cormier and Kieffer [22], who used a comparable surgical approach and protocol.

In terms of the surgery, resection of the cervical rib, first rib or hypertrophic middle scalene had no effect on the outcomes in our study. This is consistent with other published studies [18,24]. Sanders and Hammond reported a similar success rate (70%) with and without resection of the first rib [27]. Degeorges et al. reported that partial resection of the first rib was beneficial [28]. Peek et al. reported that 54% of patients had their symptoms completely resolved and 90% had clinical improvement after surgical treatment [29].

While our results differ (12% when first rib is resected versus 35% when it is not), this difference was not statistically significant. First rib resection is not universally accepted [30] as it increase the morbidity rate [19,26]. "A la carte" treatment with first rib resection if the condition is not post-traumatic and based on the intraoperative observations is a reasonable approach. Resection appears necessary in case of recurrence, if no abnormality was identified during the procedure and if vein or artery damage is identified [27].

In a 2014 Cochrane review [31] on the effectiveness of TOS treatments, only two studies were retained that provided evidence on treatment efficacy. One study of 55 patients showed that first rib resection by the Roos approach reduced the pain more than neurolysis by the supraclavicular approach [32], while the second study showed a reduction in paresthesia after Botox injection in the anterior scalene [33]. All the other studies identified were excluded because of design flaws, namely lack of randomization. Thus it is difficult to draw scientific conclusions with a sufficient level of evidence. Functional treatment, which can lead to good outcomes [34], has not been compared to surgical treatment. The possibility of sequelae and the persistence of neuropathic pain means the surgical indication should be limited to patients with significant functional disability despite well-conducted rehabilitation [22].

5. Conclusion

The majority of patients operated for non-specific TOS using the supraclavicular approach had significantly improvement after the procedure based on the QuickDASH Score. After surgery, their pain level, paresthesia, numbness and weakness in the upper limb was significantly better. Treatment of nonspecific TOS could be improved by getting a consensus on the diagnostic criteria and by comparing the effectiveness of various treatments in randomized prospective studies.

Disclosure of interest

The authors declare that they have no competing interest.

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