

Rare etiologies of thoracic outlet syndrome: A systematic review

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Abstract. Thoracic outlet syndrome (TOS) encompasses a group of symptoms resulting from the compression of neurovascular structures within the thoracic outlet. While TOS is often linked to repetitive activities, trauma, or anatomical variations, rare causes can also play a critical role in this condition. The present study systematically explored these unusual etiologies of TOS. A comprehensive search was conducted using PubMed and Google Scholar databases to identify relevant studies in the English language on rare causes of TOS. Studies addressing well-recognized causes were excluded. Data extracted from the selected studies included the authors, year of publication, number of cases, patient demographics, history of trauma or TOS, symptoms, affected side, Roos test results, misdiagnoses, the underlying cause of TOS, the type of TOS, diagnostic approach, management strategies, outcomes, follow-up details and recurrence rates. The main findings were summarized using means, ranges, frequencies and percentages. A total of 246 articles were screened, and 30 case reports involving 30 patients met the inclusion criteria. The mean age of the patients in the studies was 40.83 ± 16.93 years, with 18 males (60%) and 12 females (40%). Neurogenic TOS was identified in 17 cases (56.67%), followed by neurovascular TOS in 8 cases (26.67%), venous TOS in 3 cases (10.00%) and arterial TOS in 2 cases (6.67%). Rare causes included clavicular fracture screws, subclavius posticus muscle, osteoblastoma, lipoma, Nuss procedure, chondrosarcoma and several other unusual conditions. The complexity and

variability of rare causes of TOS underscore the importance of precise diagnostic evaluation. Recognizing these rare etiologies is essential for reducing misdiagnosis and improving patient outcomes through targeted diagnostic and treatment strategies.

Introduction

Thoracic outlet syndrome (TOS) is a group of symptoms resulting from the compression of neurovascular structures as they pass through three specific areas: The costoclavicular junction, the scalene triangle and the pectoralis minor space (1). Depending on the affected structure, TOS is divided into three primary categories as follows: neurogenic TOS (nTOS), arterial TOS (aTOS) and venous TOS (vTOS) (1).

nTOS is the most common subtype, accounting for >90% of all TOS cases. nTOS tends to occur more frequently among females and is often associated with poor posture, inadequate muscle development, or a combination of both (2). vTOS accounts for 3-5% of cases, while aTOS represents only 1%. Each subtype can be associated with traumatic, congenital, or acquired factors, with trauma being a predominant contributor in most cases (3). A typical example of a congenital TOS case is the presence of cervical ribs or an anomalous first rib. Additionally, the familial occurrence of TOS has led researchers to increasingly associate the condition with specific genetic variations (3). Traumatic cases are predominantly caused by whiplash injuries and falls, which may result in TOS either directly, through structural damage, or indirectly. Long-term compression of neurovascular structures can occur due to bone deformation, scalene muscle alterations, or cervical plexus involvement following trauma. Acquired causes are commonly linked to occupational activities, sports, or repetitive movements (3).

The diagnosis of TOS is often complicated by its varied clinical presentations, which frequently mimic other conditions and contribute to a high rate of misdiagnosis. This challenge is further exacerbated by the lack of a universally accepted diagnostic standard, resulting in a reliance on clinical judgment and inconsistent use of diagnostic tests (4).

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Rare causes of TOS are particularly vulnerable to being overlooked due to their atypical features and the generally low clinical suspicion, increasing the risk of delayed or inaccurate diagnosis. Nonetheless, these uncommon etiologies should not be disregarded, as a more in-depth understanding of these is essential for enhancing diagnostic precision and guiding appropriate management (5).

The present study aimed to systematically review the rare causes of TOS to highlight their clinical relevance and promote improved recognition and diagnosis in practice.

Data and methods

Study design. The present systematic review was conducted following the protocols established by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.

Data sources and search strategy. A comprehensive search was conducted using the PubMed and Google Scholar databases to identify relevant studies in the English language on rare causes of TOS. The search utilized the following terms: 'thoracic outlet syndrome' with 'case reports', 'rare cause', 'unusual', 'uncommon', 'infrequent', 'odd', and 'sporadic'. No publication date restrictions were applied.

Eligibility criteria. The present study included all reported articles on rare causes of TOS, excluding those that: i) Resulted from well-recognized structural or anatomical abnormalities such as cervical ribs; ii) involved functional or dynamic causes; iii) were associated with nonunion or malunion of the clavicle; iv) were not published in the English language; v) were available only as abstracts; or vi) lacked proper peer review as determined by cross-checking the journal against recognized lists of predatory publications (6).

Study selection and data extraction. Before conducting a full-text eligibility assessment, the titles and abstracts of the included studies were initially screened. Data were extracted from the selected studies, including the authors, year of publication, number of cases, age, sex, occupation, history of trauma, TOS history, symptoms, affected side, results of the provocative tests, misdiagnosis, cause of TOS, TOS type, diagnostic approach, management, outcome, follow-up and recurrence.

Data presentation. The data were structured in an Excel spreadsheet (Microsoft Excel, 2021) and analyzed qualitatively using the Statistical Package for the Social Sciences (SPSS, version 27.0). The main findings are reported as the mean, ranges, frequencies and percentages.

Results

A total of 246 articles were initially identified from the searched databases. Of these, 33 articles were removed prior to screening as they were duplicates, published only as abstracts, or written in languages other than English. In the first screening phase, 170 out of 213 articles were removed as they did not meet the inclusion criteria. Full-text screening

was performed on the remaining 43 articles; however, 11 articles could not be retrieved and were excluded. After assessing the eligibility of the remaining 32 articles, two articles were excluded due to an inadequate peer review process, as determined by cross-referencing the journals with recognized lists of predatory publications (Fig. 1). Finally, 30 case reports (5,7-35), including 30 cases, met the inclusion criteria.

The mean age of the patients in the included studies was 40.83±16.93 years, with ages ranging from 16 to 80 years. There were 18 males (60%) and 12 females (40%). Occupational data were unavailable for 25 patients (83.33%). Additionally, 25 patients (83.33%) reported no history of prior non-surgical trauma (Table I).

Clinically, pain was the most frequently reported symptom, present in 21 patients (70%), followed by paresthesia in 19 (63.33%) patients, weakness in 10 (33.33%) patients and numbness in 7 (23.33%) patients. The left side was affected in 15 cases (50%), the right in 10 cases (33.33%), and bilateral involvement was noted in 2 cases (6.67%).

The initial diagnosis of TOS was established using CT scans and MRI in 10 cases each (33.33%), and clinical evaluation alone in 6 cases (20%). The types of TOS reported included nTOS in 17 patients (56.67%), neurovascular in 8 (26.67%), vTOS in 3 (10%) and aTOS in 2 patients (6.67%) (Table II).

The most frequently reported rare etiologies were clavicular fracture fixation screws and the subclavius posticus muscle, each accounting for 3 cases (10%). Osteoblastoma, lipoma and complications from the Nuss procedure were each identified in 2 cases (6.67%). The remaining 18 cases (60%) involved various other rare causes (Table III).

Surgical intervention was performed in 22 patients (73.33%), while conservative management was employed in 7 patients (23.33%). The reported outcomes indicated that 21 patients (70%) achieved complete symptom resolution, 8 (26.66%) achieved partial improvement, and outcome data were unavailable for 1 patient (3.33%).

The follow-up duration was <6 months in 15 patients (50%), between 7 months and 2 years in 9 patients (30%) and >2 years in 4 patients (13.33%). No cases of recurrence were reported during follow-up (Table IV).

Discussion

The superior thoracic outlet is an anatomical region where the brachial plexus, subclavian artery, and subclavian vein pass. It is located between the anterior and middle scalene muscles, above the first rib, behind the clavicle, and lateral to the sternal manubrium (36). The scalene triangle is the primary site for compressions of the subclavian artery and brachial plexus. By contrast, venous compressions primarily occur in the costoclavicular space (12).

The epidemiology of TOS is unclear, largely due to the lack of standardized diagnostic criteria. Incidence estimates range from 3 to 80 per 1,000 individuals, with a higher prevalence among females aged 20-50 years (37,38). In the present systematic review, 60% of the patients in the included studies were male, with a mean age of 40.83 years, ranging from 16 to 80 years.

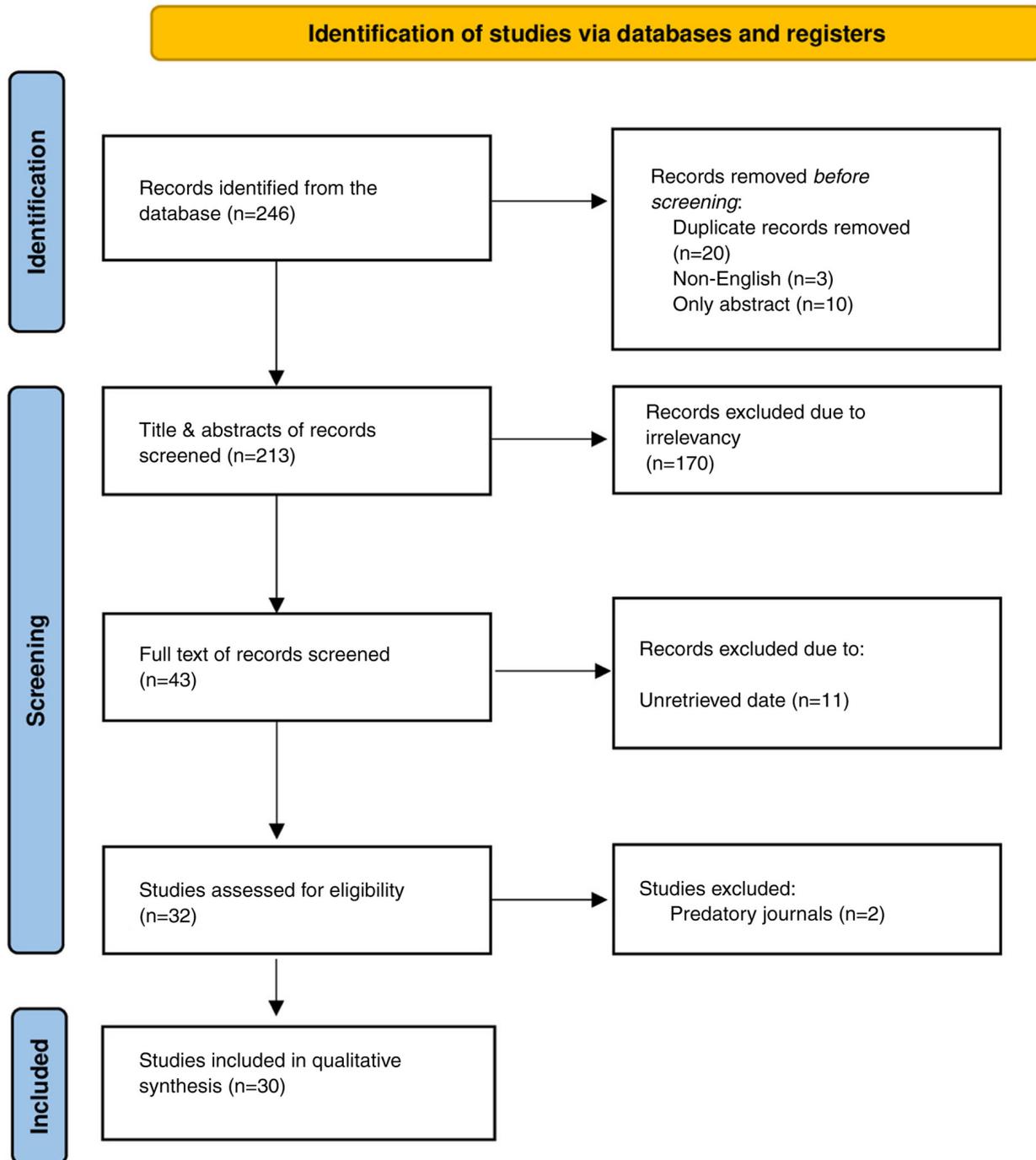


Figure 1. PRISMA flow diagram illustrating the study selection process.

True nTOS presents with pain, paresthesia and weakness in areas served by the affected nerve roots. vTOS, or Paget-Schroetter syndrome, causes sudden upper limb swelling, cyanosis, venous engorgement and pain, often triggered by thrombus in the subclavian vein. aTOS, though rarest, is the most severe, marked by pain, pallor, reduced radial pulse with arm movement, fatigue and signs of limb ischemia due to embolization (38). In the present systematic review, 56.67% of the patients from the included studies had nTOS, 26.67% had neurovascular TOS, 10% had vTOS, and 6.67% had aTOS. Pain was the most common symptom, reported by 70% of patients, followed by paresthesia, which was reported by 63.33% of patients.

TOS diagnosis begins with a clinical evaluation, including a history and physical examination to identify symptoms, such as upper limb pain or numbness, suggesting neurovascular compression (39). Provocative tests such as Adson's and Roos tests help assess vascular and neurological involvement (40). In the present systematic review, Roos test results were reported in only 3 cases, with 2 of these exhibiting positive results. Ultrasound is valuable in diagnosing nTOS due to its dynamic imaging, enabling real-time visualization of structures during movements that often trigger symptoms, such as overhead reaching or head turning. Magnetic resonance neurography helps detect anatomical

Table I. Information from the included case reports.

First author	Year of publication	No. of cases	Age (years)	Sex	Occupation	Hx of non-surgical trauma		Hx of TOS	Common symptoms in the upper limb				Dominant hand	Affected side	(Refs.)
						Clavicular fracture	Left humerus neck fracture		Paresthesia	Pain	Numbness	Weakness			
Hamouri	2021	1	23	Female	N/A	No	No	No	Yes	No	No	N/A	Left	(5)	
Hussain	2024	1	67	Male	N/A	Clavicular fracture		No	Yes	No	No	N/A	Left	(7)	
Kakamad	2024	1	45	Female	N/A	No	No	No	Yes	No	No	N/A	N/A	(8)	
Philp	2024	1	36	Female	Professional golfer	No	No	No	Yes	No	No	N/A	Left	(9)	
Kimura	2023	1	40	Male	golf player	No	No	No	Yes	Yes	No	N/A	Left	(10)	
Lee	2023	1	71	Male	Carpenter	Left humerus neck fracture		No	Yes	No	Yes	Right	Left	(11)	
Mansouri	2023	1	41	Male	Mechanic	No	No	No	No	No	No	N/A	Left	(12)	
Singh	2023	1	21	Female	N/A	No	No	Yes	Yes	No	No	N/A	Right	(13)	
Fernandes	2022	1	15	Male	N/A	No	No	No	Yes	No	Yes	N/A	Right	(14)	
Ohida	2021	1	50	Female	N/A	No	No	No	Yes	No	Yes	N/A	Left	(15)	
Fleet	2020	1	32	Female	N/A	No	No	No	Yes	No	No	N/A	Left	(16)	
McAleese	2020	1	23	Female	N/A	SCJ dislocation		No	Yes	No	Yes	N/A	N/A	(17)	
Tangpiroontham	2020	1	30	Female	N/A	No	No	No	No	Yes	Yes	N/A	Right	(18)	
Liu	2019	1	80	Male	N/A	No	No	No	No	No	No	N/A	Right	(19)	
Sul	2019	1	45	Male	N/A	No	No	No	Yes	No	No	N/A	Left	(20)	
Wang	2019	1	66	Male	N/A	No	No	No	No	Yes	Yes	N/A	Right	(21)	
Zhang	2018	1	27	Male	N/A	No	No	No	Yes	Yes	Yes	N/A	Right	(22)	
Vaidya	2017	1	24	Female	N/A	No	No	No	No	Yes	No	N/A	Bilateral	(23)	
Medina	2016	1	17	Male	N/A	No	No	No	No	Yes	Yes	N/A	Left	(24)	
Patel	2016	1	55	Male	N/A	No	No	No	Yes	No	No	N/A	Right	(25)	
Sergeant	2016	1	70	Male	N/A	No	No	No	Yes	No	No	N/A	N/A	(26)	
Zampieri	2016	1	41	Female	N/A	No	No	No	Yes	No	No	N/A	Left	(27)	
Mueller	2015	1	32	Male	N/A	No	No	No	No	No	Yes	N/A	Left	(28)	
Smayra	2013	1	30	Female	N/A	No	No	No	No	Yes	No	N/A	Left	(29)	
Jennings	2012	1	45	Male	N/A	Clavicular fracture		No	Yes	No	No	Right	Right	(30)	
Skedros	2010	1	38	Male	Manual laborer	Clavicular fracture		No	No	Yes	Yes	Right	Right	(31)	
Singh	2009	1	50	Male	Machine operator	No	No	No	Yes	No	Yes	Right	Bilateral	(32)	
Nakazawa	2005	1	34	Male	N/A	No	No	No	Yes	No	No	N/A	Right	(33)	
Chen	1999	1	44	Female	Secretary	No	No	No	No	Yes	No	Right	Right	(34)	
Gehman	1998	1	33	Male	N/A	No	No	Yes	Yes	No	No	N/A	Left	(35)	

N/A, non-available; SCJ, posterior sternoclavicular joint; Hx, history; TOS, thoracic outlet syndrome.

Table II. Overview of the clinical data from the included studies.

First author	MisDx	Dx with TOS by	Cause of TOS	Type of TOS	Type of management	Outcome	Follow-up (month)	Recurrence	(Refs.)
Hamouri	N/A	CT scan	Rib osteoblastoma	Neurogenic	Surgical management	Complete symptom relief	24	No	(5)
Hussain	N/A	CTA + Doppler US	Clavicular fracture fixation screw	Arterial	Surgical management	Complete symptom relief	1.5	No	(7)
Kakamad	N/A	CT scan + Physical exam	Mediastinal mass	Neurogenic	Surgical management	Complete symptom relief	2	No	(8)
Philp	Adhesive capsulitis	Physical exam	Trapezius muscle dystonia	Neurogenic	Conservative management	Improvement in symptoms	12	No	(9)
Kimura	N/A	CT scan + MRI	Anomalous muscle	Neurogenic	Surgical management	Complete symptom relief	36	No	(10)
Lee	N/A	MRI	Subclavius posticus muscle	Neurogenic	Surgical management	Complete symptom relief	6	No	(11)
Mansouri	Arthritis of the sternoclavicular joint	CT scan	Manubrium sterni chondrosarcoma	Venous	Surgical management	Complete symptom relief	12	No	(12)
Singh	N/A	X. ray	Cervical rib osteoblastoma	Neurogenic	Multimodal therapy	Improvement in symptoms	N/A	No	(13)
Fernandes	N/A	Physical exam + Doppler US	Nuss procedure	Neurovascular	Conservative management	Complete symptom relief	7	No	(14)
Ohida	N/A	CT scan + Duplex US	SAPHO syndrome	Neurovascular	Surgical management	Complete symptom relief	2	No	(15)
Fleet	N/A	MRI	Brachial neuritis	Neurogenic	Surgical management	Improvement in symptoms	30	No	(16)
McAleese	N/A	CT scan	Posterior displacement of a Salter-Harris Type II fracture-dislocation	Neurovascular	Conservative management	Complete symptom relief	36	No	(17)
Tangpiroontham	N/A	EMG + MRI	Axillary breast implantation	Neurovascular	Surgical management	Improvement in symptoms	6	No	(18)
Liu	N/A	CTA + US	Stent implantation for AVM	Venous	Surgical management	Complete symptom relief	6	No	(19)
Sul	N/A	CT scan + X. ray	Lipoma in the pectoralis minor space	Neurovascular	Surgical management	Complete symptom relief	12	No	(20)
Wang	N/A	Physical exam + MRI	Surgery for cervical spondylotic myelopathy	Neurogenic	Conservative management	Improvement in symptoms	12	No	(21)

Table II. Continued.

First author	MisDx	Dx with TOS by	Cause of TOS	Type of TOS	Type of management	Outcome	Follow-up (month)	Recurrence (Refs.)
Zhang	N/A	EMG + CTA	Nuss procedure	Neurovascular	Conservative management	Improvement in symptoms	12	No (22)
Vaidya	N/A	Physical exam + MRI	Lymphatic cyst	Neurogenic	Surgical management	N/A	N/A	No (23)
Medina	N/A	CT scan + MRI	Aneurysmal bone cyst	Neurogenic	Surgical management	Complete symptom relief	<1	No (24)
Patel	N/A	CT scan + X-ray	Chondrosarcoma	Neurogenic	Surgical management	Complete symptom relief	21	No (25)
Sergeant	N/A	CT scan	Subpectoral lipoma	Neurogenic	Surgical management	Complete symptom relief	1	No (26)
Zampieri	N/A	Doppler US	Chylous cyst	Venous	Surgical management	Complete symptom relief	3	No (27)
Mueller	Carpal tunnel syndrome	MRI	Subclavius posticus muscle	Neurogenic	Conservative management	Complete symptom relief	6	No (28)
Smayra	N/A	CTA + Doppler US	Subclavius posticus muscle	Neurovascular	Surgical management	Complete symptom relief	2	No (29)
Jennings	N/A	Arteriogram	Clavicular fracture fixation screw	Arterial	Surgical management	Improvement in symptoms	3	No (30)
Skedros	N/A	Venogram	Clavicular fracture fixation screw	Neurogenic	Surgical management	Improvement in symptoms	48	No (31)
Singh	Cervical spondylosis	NCS	Bilateral coracoclavicular joints	Neurogenic	Conservative management	Complete symptom relief	18	No (32)
Nakazawa	N/A	MRI	Neurilemmoma in the pectoralis minor space	Neurogenic	Surgical management	Complete symptom relief	<1	No (33)
Chen	Fibromyalgia	Physical exam	Intraosseous ganglion cyst	Neurogenic	Surgical management	Complete symptom relief	5	No (34)
Gehman	N/A	MRI	Desmoid tumor	Neurovascular	Surgical management	Complete symptom relief	6	No (35)

N/A, not available; MisDx, misdiagnosis; Dx, diagnosis; TOS, thoracic outlet syndrome; CTA, computed tomography angiography; US, ultrasound; MRI, magnetic resonance imaging; EMG, electromyography; NCS, nerve conduction study; SAPHO, synovitis, acne, pustulosis, hyperostosis and osteitis; AVM, arteriovenous malformation.

Table III. Rare causes of thoracic outlet syndrome and their frequency.

Causes	Frequency and percentage (%)
Clavicular fracture fixation screw	3 (10.00)
Subclavius posticus muscle	3 (10.00)
Osteoblastoma	2 (6.67)
Pectoralis minor space or subpectoral lipoma	2 (6.67)
Nuss procedure	2 (6.67)
Aneurysmal bone cyst	1 (3.33)
Anomalous muscle	1 (3.33)
Axillary breast implantation	1 (3.33)
Bilateral coracoclavicular joints	1 (3.33)
Brachial neuritis	1 (3.33)
Chondrosarcoma	1 (3.33)
Chylous cyst	1 (3.33)
Desmoid tumor	1 (3.33)
Trapezius muscle dystonia	1 (3.33)
Intraosseous ganglion cyst	1 (3.33)
Lymphatic cyst	1 (3.33)
Manubrium sterni chondrosarcoma	1 (3.33)
Mediastinal mass	1 (3.33)
Neurilemmoma in the pectoralis minor space	1 (3.33)
Posterior displacement of a Salter-Harris	1 (3.33)
Type II fracture-dislocation	
SAPHO syndrome	1 (3.33)
Stent implantation for AVM	1 (3.33)
Surgery for cervical spondylotic myelopathy	1 (3.33)

SAPHO, synovitis, acne, pustulosis, hyperostosis and osteitis; AVM, arteriovenous malformation.

abnormalities, while electrodiagnostic tests reveal nerve dysfunction (41,42).

Various etiologies contribute to TOS, each typically associated with a specific type of TOS. Certain conditions, such as multiple exostoses, can cause a combination of nTOS and vTOS (12). Anatomical variations, such as the presence of a cervical rib or abnormalities in the scalene muscles' thickness, insertion, or structure and fibroligamentous tissues, may also contribute to TOS. Additionally, abnormal widening of the sternal (>20 mm) and vertebral (>15 mm) ends of the first rib is a potential cause (12). Traumatic origins, including fractures of the clavicle or first rib and cervical trauma (e.g., whiplash injury), are the most common causes of neurological symptoms in TOS. Moreover, vTOS can develop following acute or chronic compression of the subclavian vein due to intense upper limb strain or compression from a hypertrophic bone callus after a fracture (12). However, familial predisposition can influence the manifestation of TOS. A previous study reported a rare case involving multiple family members diagnosed with TOS, indicating a potential genetic link despite the absence of identifiable syndromes or mutations in known genes, such as the HOX gene family (43). These findings

indicate the possibility of polygenic inheritance, with strong phenotypic penetrance observed in affected families. While specific genetic mutations remain unidentified, variations in HOX gene expression are proposed as potential contributors to the anatomical anomalies associated with TOS (43).

While the typical causes of TOS are well understood, several rare causes merit attention. One such cause is anomalous muscle formations, which can compress the brachial plexus or subclavian vessels. Kimura *et al* (10) reported a case involving a 40-year-old male carpenter presenting with numbness in his left upper extremity, initially misdiagnosed as cervical spondylotic radiculopathy. Further imaging identified an anomalous muscle compressing the left brachial plexus, which was determined to be the source of his symptoms (10). However, the case study by Muellner *et al* (28) highlighted a 32-year-old male who experienced hand weakness. MRI imaging revealed the presence of the subclavius posticus muscle, which caused significant narrowing of the costoclavicular space and contributed to intermittent brachial plexus compression. While this condition is relatively rare, with only a few documented cases, it emphasizes the importance of considering the subclavius posticus muscle as a potential underlying factor in nTOS (28). The present study revealed that the subclavius posticus muscle was responsible for causing 3 cases of TOS. These findings underscore the need to consider anomalous muscle formations when diagnosing TOS, particularly in patients with unclear or atypical presentations.

Recent studies have indicated that excessive muscle development in the trapezius region can elevate the distal clavicle and narrow the thoracic outlet space. The case report by Philp *et al* (9) highlighted significant symptom improvement in a patient with nTOS following botulinum toxin injections to reduce trapezius muscle tone. This finding suggests that muscular imbalances may play a more critical role in the pathophysiology of TOS than previously recognized.

Another rare cause of TOS involves space-occupying lesions, which can compress the neurovascular bundle and produce TOS-like symptoms. Sul *et al* (20) reported a case involving a 45-year-old male who presented with paresthesia and tingling in his left arm, attributed to a large lipoma compressing the lower trunk of the brachial plexus. The mass extended from the pectoralis minor space into the costoclavicular space, resulting in significant neurovascular compression (20). However, Kakamad *et al* (8) reported the case of a 45-year-old female patient who experienced persistent pain and numbness in her left upper limb. Imaging revealed a soft tissue mass in the superior mediastinum, which encased the left subclavian artery and partially involved the left innominate vein. Despite initial conservative treatments and decompression surgery, the symptoms of the patient worsened until a median sternotomy was performed for total resection of the mass, ultimately resulting in complete symptom relief (8).

Post-surgical changes can also give rise to rare forms of TOS. In the case reported by Hussain *et al* (7), TOS was caused by an excessively long fixation screw from a previous clavicular fracture surgery, which impinged on both the left subclavian artery and the brachial plexus. The improper screw length resulted in chronic irritation and damage to the arterial wall, leading to thrombosis and limb-threatening ischemia, which manifested seven years after surgery. The authors of

Table IV. Overview of the key characteristics of the included case studies.

Parameters	Value/frequency
Age, mean ± SD	40.83±16.93
Sex, n (%)	
Male	18 (60.00%)
Female	12 (40.00%)
Occupation, n (%)	
Professional golf player	1 (3.33%)
Mechanic	1 (3.33%)
Manual laborer	1 (3.33%)
Machine Operator	1 (3.33%)
Secretary	1 (3.33%)
N/A	25 (83.33%)
History of non-surgical trauma, n (%)	
Clavicular fracture	3 (10.00%)
Left humerus neck fracture	1 (3.33%)
Posterior displacement at the sternoclavicular joint	1 (3.33%)
None	25 (83.33%)
Upper limb common symptoms, n (%)	
Pain	21 (70.00%)
Paresthesia	19 (63.33%)
Weakness	10 (33.33%)
Numbness	7 (23.33%)
Affected side, n (%)	
Left	15 (50.00%)
Right	10 (33.33%)
Bilateral	2 (6.67%)
N/A	3 (10.00%)
Misdiagnosis, n (%)	
Adhesive capsulitis	1 (3.33%)
Arthritis of the sternoclavicular joint	1 (3.33%)
Carpal tunnel syndrome	1 (3.33%)
Cervical spondylosis	1 (3.33%)
Fibromyalgia	1 (3.33%)
Upper extremity pain syndrome	1 (3.33%)
None	24 (80.00%)
Roos test, n (%)	
Positive	2 (6.67%)
Negative	1 (3.33%)
N/A	27 (90.00%)
Type of TOS, n (%)	
Neurogenic	17 (56.67%)
Neurovascular	8 (26.67%)
Venous	3 (10.00%)
Arterial	2 (6.67%)
TOS initially diagnosed by, n (%) ^a	
CT scan	10 (33.33%)
MRI	10 (33.33%)
Clinical evaluation	6 (20.00%)
Doppler US	4 (13.33%)
CTA	4 (13.33%)

Table IV. Continued.

Parameters	Value/frequency
X-ray	3 (10.00%)
EMG	2 (6.67%)
Duplex US	1 (3.33%)
US	1 (3.33%)
Arteriogram	1 (3.33%)
Venogram	1 (3.33%)
NCS	1 (3.33%)
Type of management, n (%)	
Surgical management	22 (73.33%)
Conservative management	7 (23.33%)
Multimodal therapy	1 (3.33%)
Outcome, n (%)	
Complete symptom relief	21 (70.00%)
Improvement in symptoms	8 (26.66%)
N/A	1 (3.33%)
Follow-up duration, n (%)	
<1-6 months	15 (50.00%)
7 months to 2 years	9 (30.00%)
>2 years	4 (13.33%)
N/A	2 (6.67%)
Recurrence, n (%)	
No	30 (100.00%)
Yes	0 (0.00%)

^aIn some cases, TOS was initially diagnosed using two diagnostic approaches. N/A, not available; TOS, thoracic outlet syndrome; CT scan, computed tomography scan; MRI, magnetic resonance imaging; US, ultrasound; NCS, nerve conduction study; SD, standard deviation; CTA, computed tomography angiography; EMG, electromyography.

that study emphasized that such complications can develop insidiously and stress the importance of vigilant post-operative monitoring of patients following clavicular fixation to prevent serious vascular issues (7). However, Fernandes *et al* (14) reported a rare case of TOS in a 15-year-old boy following the Nuss procedure for pectus excavatum. At 2 weeks post-surgery, he developed right-hand paresthesia, weakness and coldness in the arm due to the compression of the subclavian artery and brachial plexus, likely resulting from the structural changes in the thoracic cavity caused by the surgery (14). In the present study, 3 cases of TOS were found to be attributed to clavicular fracture fixation screws, and two cases were associated with the Nuss procedure. These findings further highlight the importance of considering post-surgical complications in the differential diagnosis of TOS, particularly in patients with a history of clavicular surgery or thoracic procedures.

Diagnosing rare forms of TOS poses significant challenges due to their atypical presentations and the overlapping symptoms with more common neurological or musculoskeletal conditions. In numerous cases, misdiagnosis or delayed diagnosis occurs, particularly when symptoms mimic cervical radiculopathy, adhesive capsulitis, or brachial neuritis, as

highlighted by Fleet *et al* (16), where nTOS was initially overlooked in favor of brachial neuritis. Imaging plays a critical role in diagnosing rare TOS etiologies. MRI with provocative positioning can identify dynamic compressions, as seen in patients with nTOS secondary to anomalous muscles or anatomical variations (18). In the present systematic review, it was found that the diagnostic methods used for TOS varied. CT scans and MRIs each identified 33.33% of cases. Clinical diagnosis accounted for 20.00%, while Doppler ultrasound and computed tomography angiography each diagnosed 13.33% of cases. The other diagnostic methods were used less frequently.

Misdiagnoses are common when rare etiologies are not considered, leading to prolonged symptoms and functional impairment. In the case reported by Philp *et al* (9), trapezius hypertonicity and hypertrophy were unrecognized contributors to nTOS until botulinum toxin injection into the trapezius led to significant symptom improvement.

Treatment for rare cases of TOS is highly individualized. Surgical intervention remains the cornerstone for cases involving structural abnormalities or space-occupying lesions (13,18). Nonetheless, conservative management can be successful in selected cases. Fernandes *et al* (14) reported complete recovery following rehabilitation and nerve-nourishing therapy in a patient with Nuss procedure-induced TOS, avoiding the need for bar removal. In the present systematic review, in the included studies, surgical intervention was performed in 22 patients (73.33%), while 7 patients (23.33%) were managed conservatively. The reported outcomes indicated that 70% of patients achieved complete symptom resolution, while 26.66% experienced partial improvement.

Further research is warranted to focus on elucidating the genetic and anatomical factors that predispose individuals to rare forms of TOS, including the potential role of polygenic inheritance and variations in gene expression.

In conclusion, the complexity and variability of rare causes of TOS underscore the importance of precise diagnostic evaluation. These rare etiologies can include structural anomalies, anomalous muscles, space-occupying lesions, and iatrogenic causes such as the Nuss procedure. Recognizing these rare etiologies is essential for reducing misdiagnosis and improving patient outcomes through targeted diagnostic and treatment strategies.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

FHK, AKG and SKA were major contributors to the conception of the study, as well as to the literature search for related

studies. HAN, BAA and HSN were involved in the literature review, IN the writing of the manuscript, in the design of the study and in the interpretation of the data obtained from the literature. NSS, SHT, LJM, YNA, CSO, AHA, ASH and LAS were involved in the literature review, in the design of the study, and in the critical revision of the manuscript. BAA and FHK confirm the authenticity of all the raw data. All authors have read and approved the final version of the manuscript.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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