From the Southern Association for Vascular Surgery

Boney abnormalities cause arterial, venous, and/or neurogenic thoracic outlet syndrome

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ABSTRACT

Background: Thoracic outlet syndrome (TOS) is a rare condition caused by compression of the neurovascular structures within the thoracic outlet. Different classifications of TOS exist depending on the neurovascular structure being compressed: neurogenic, venous, or arterial. Any of these forms can present independently or coexist with one other. TOS symptoms are sometimes precipitated by the presence of boney abnormalities that often require surgical intervention for ultimate resolution. This retrospective review will examine the presentations and outcomes of patients with TOS whose cause was a boney abnormality.

Methods: A total of 73 patients who underwent thoracic outlet surgery between 2016 and 2021 were retrospectively reviewed via electronic medical records. Twelve (16%) patients demonstrated boney abnormalities on presentation causing their symptoms. The patients with boney abnormalities were analyzed based on venous, arterial, or neurogenic TOS diagnosis.

Results: Of the 12 patients with boney abnormalities, 5 were classified as venous TOS, 6 patients as neurogenic TOS, and 1 as arterial TOS. The boney abnormalities were as follows: venous TOS: three clavicular fractures, one nonfused congenital clavicle, and one residual rib; neurogenic TOS: three fractured first ribs, one fractured clavicle, and two cervical ribs; and arterial TOS: fused first and second rib with bilateral cervical ribs and arterial compression. Postoperatively, there were no artery, vein, or nerve injuries. Five patients had a pneumothorax treated over night with a chest tube, and one patient had a superficial wound infection. The median hospital stay was 1 day. All patients completed physical therapy after surgery. All patients have symptom resolution at follow-up.

Conclusions: Patients with boney abnormalities constitute about one-fifth of patients who can present with all three forms of TOS: neurogenic, arterial, and venous, and some will have more than one of these presentations. Results in patients undergoing surgery with boney abnormalities causing thoracic outlet syndrome are excellent with symptom resolution and without substantial complications. (J Vasc Surg Cases Innov Tech 2023;9:1-5.)

Keywords: Thoracic outlet syndrome; Arterial; Venous; Neurogenic; Boney abnormality; Cervical ribs; Trauma

Thoracic outlet syndrome (TOS) is a complex disease that affects 3 to 80 per 1000 of the population. TOS arises because of compression of the thoracic outlet, a space that includes the interscalene triangle, the costoclavicular space, and the retropectoralis space.[1,2] Narrowing of the costoclavicular space can compress one or more neurovascular structures that include the brachial plexus, subclavian artery, or subclavian vein. Compression can be caused by chronic repetitive motion (CRM) and/or a traumatic injury/accident, all of which can inflame the anterior scalene muscle, drawing the first rib up superiorly and narrowing the thoracic outlet. Depending on the structure that is compressed, the syndrome can present as arterial (ATOS, <1% of cases), venous (VTOS, 3%-5%), or neurogenic (NTOS, >90%).[3]

In addition to CRM and trauma, boney abnormalities can also lead to the development of TOS. Boney abnormalities can impinge in the thoracic outlet resulting in TOS symptoms. Boney abnormalities include the presence of a residual or rudimentary ribs, clavicular anomalies, fused and fractured ribs, or supernumerary cervical ribs that are typically unilateral but can sometimes be bilateral.[4,5]

These boney abnormalities compress the neurovascular bundle through a variety of mechanisms. The thoracic outlet is directly posterior to the clavicle, and compromise of clavicular function due to neck/chest trauma can result in irritation of the brachial plexus and/or compression of the vascular structures within the outlet.[2] In addition, a residual rib from previous intervention can lead to recurrent TOS symptoms as

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scar tissue and fibrous cartilage anchor onto the remaining rib remnant.

Complete surgical resection of the boney abnormality is essential for TOS symptom resolution. Without removal, scar tissue and fibrous cartilage can prevent mobility and flexibility within the space, leading to recurrent symptoms.[6] Current research appropriately attributes boney abnormalities to TOS, but the definition of what constitutes a boney abnormality, its proper identification, and surgical removal is unclear and decentralized. In this article, we describe a cohort of patients with boney abnormalities that caused TOS and their outcomes after resection of the boney abnormality in addition to a first rib resection and anterior scalenectomy (FRRAS).

METHODS

Study participants. In 2020, Atrium Health Wake Forest Baptist Department of Vascular and Endovascular Surgery received institutional review board approval to develop and maintain an electronic database of all patients who underwent FRRAS for TOS between 2016 and 2021. A total of 73 patients were retrospectively reviewed, and all provided informed consent for inclusion in the database.

Data collection. Before surgical intervention, patients were physically examined, symptomatology recorded, and diagnostic modalities such as chest radiograph, duplex ultrasound imaging, and occasional computed tomography were used.

Demographic and clinical examination records were obtained via electronic medical records. Demographic factors included sex, age, and history of physical accidents or CRM. Clinical factors included length of symptoms before initial visit, time between initial visit and surgery date, length of follow-up, length of anticoagulation use, and presence of a boney abnormality detected by a chest radiograph. Boney abnormalities were defined as a rudimentary rib, residual rib, fractured rib, fused rib, clavicular fracture, congenital clavicular anomaly, and the presence of a cervical rib.

Two weeks after operation, all patients began a prescribed physical therapy regimen of 4 to 12 weeks. All patients completed physical therapy. Patients with VTOS and ATOS obtained a venogram or angiogram to determine ipsilateral subclavian vein/artery patency. At each follow-up visit, patients with VTOS and ATOS received a duplex scan. Patients with patent vessels were taken off oral anticoagulation.

RESULTS

Of the 73 patients diagnosed with TOS, 12 patients demonstrated boney abnormalities on presentation causing and/or elevating the severity of their symptoms and opted for an FRRAS. All 12 operations were performed by two surgeons within our vascular practice.

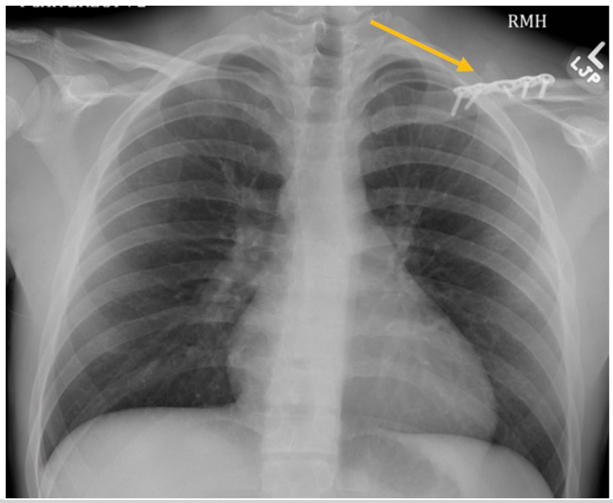

Fig 1. Fractured clavicle (repaired) chest radiograph.

Eleven patients were of the first surgeon who operated via the transaxillary approach, and one patient was of the second surgeon who operated via the infraclavicular approach. All types of boney abnormalities were identified within this cohort, except for rudimentary ribs (Supplementary Table I, online only). Of the 12 patients with boney abnormalities, 5 were classified with VTOS, 6 patients with NTOS, and 1 with ATOS. Eleven patients received a transaxillary FRRAS and one patient received an infraclavicular FRRAS.

Within the VTOS group, five patients presented with venous thrombosis and one had concomitant neurogenic symptoms. There were three males and two females with a median age of 30 years (range, 16-37 years). The boney abnormalities in this group included three clavicular fractures (Fig 1), one nonfused congenital clavicle, and one residual rib (Figs 2 and 3). Four patients underwent a transaxillary FRRAS followed by a venogram 2 weeks later. One patient underwent a simultaneous infraclavicular FRRAS and venogram. Three patients had angioplasty and one patient had thrombolysis followed by angioplasty. All five patients had patent veins at a median of 4 months (range, 1.6-6.2 months). The follow-up lengths of each patient were 25, 12, 6, 6, and 2 months.

Six patients presented with NTOS and five had evidence of arterial compression on arm abduction. Three patients with NTOS had a fractured first rib (Fig 4), one had a fractured clavicle (Fig 1), and two had cervical ribs (Fig 5). Three patients were male and three female, with a median age of 18 years (range, 14-60 years). All patients underwent transaxillary FRRAS. The two patients with evidence of cervical ribs also underwent transaxillary removal of both the cervical and first rib. The follow-up lengths of each patient were 8.5, 3, 2, 0.5, 0.5, and 0.5 months.

Fig 2. Residual rib surgically removed.

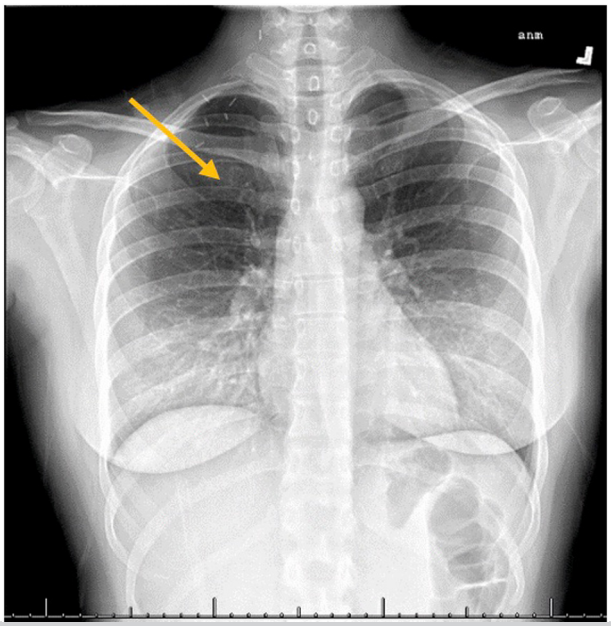

Fig 3. Residual rib chest radiograph.

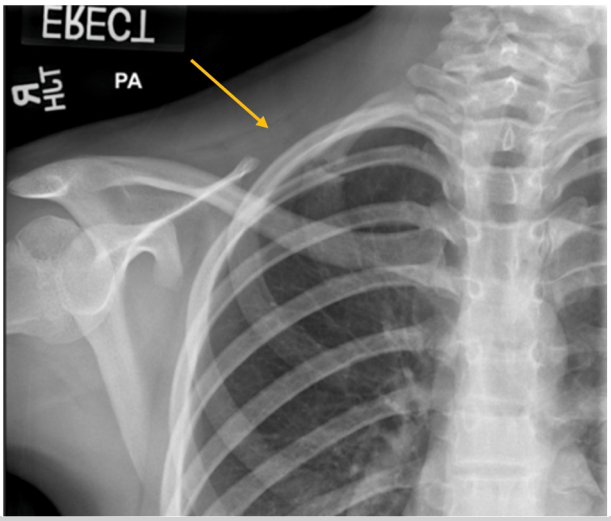

Fig 4. Fractured first rib chest radiograph.

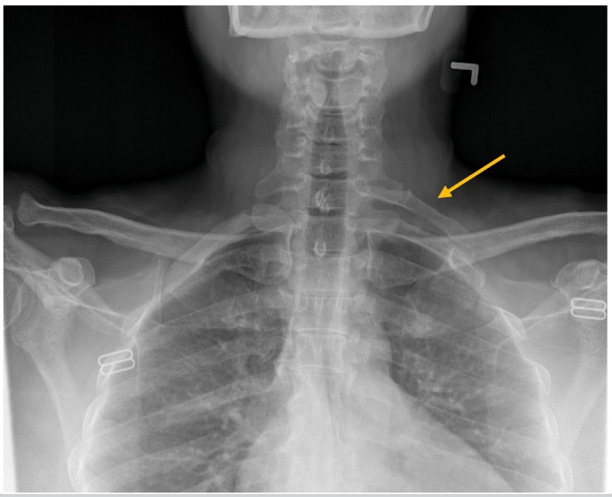

Fig 5. Cervical rib chest radiograph.

One 38-year-old woman patient with ATOS presented with a fused right first and second rib and bilateral cervical rib with arterial compression and embolization. She underwent a transaxillary FRRAS to remove the fused right first and second rib as well as the right cervical rib (Fig 6). An angiogram was performed 2 weeks later that demonstrated a patent subclavian artery. She remained on oral anticoagulation for 6.6 months to treat her distal embolization. She had complete resolution of symptoms at a follow-up length of 6.6 months.

Postoperatively, there were no artery, vein, or nerve injuries. Seven patients had a pneumothorax treated overnight with a chest tube. More pneumothoraces are seen in patients with boney abnormalities due to the inflammation and need to do more resection to remove the boney abnormality. One patient had a superficial wound infection. The median hospital stay was 1 day.

Two weeks after operation, all patients began physical therapy, which was completed 4 to 12 weeks later. Results are summarized in Supplementary Table II (online only).

DISCUSSION

Patients with boney abnormalities can present with any of the three forms of TOS. Identification of the boney abnormality in addition to correct, timely diagnosis of TOS is essential for symptom resolution. However, because of the rarity of boney abnormalities within an already rare disease, there is continued debate on the best treatment plan.

Fractured ribs. Fractures of the first rib can cause irritation, scarring, and increased fibrosis of the thoracic outlet space, resulting in irritation of the brachial plexus and vasculature. Very few fractured first ribs leading to TOS

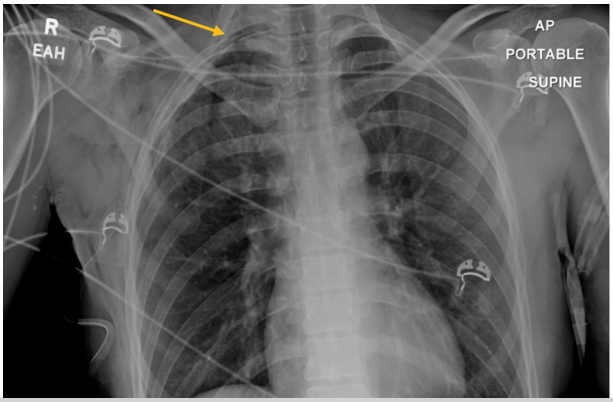

Fig 6. Fused first and second right rib and bilateral cervical rib chest radiograph.

have been reported. Terabayashi et al[7] and Mirza and Duncan[8] reported separate cases of a fractured first rib in a young athlete diagnosed with TOS, which required subsequent removal of the rib. Both case reports concluded that the surgical removal of the fractured first rib was both a safe and effective treatment.[7,8] As seen in our results, our patient in the NTOS group with a fractured first rib similarly benefited from FRRAS with no complications and complete symptom resolution (Fig 4).

Fused ribs. Fused ribs are rarely seen in TOS diagnosis. A case report on three patients described fusion of the cervical rib to the second rib, with the first rib missing.[9] All three patients underwent transaxillary cervical and second rib resection and had symptom resolution.[9] In our cohort, one patient presented with a congenital synostosis of the first and second rib, along with bilateral cervical ribs. Resection of these fused ribs provided an excellent surgical outcome for our patient with ATOS, resolving her symptoms and restoring a patent subclavian artery, further supporting the claim by Hines et al[9] that surgical resection of this anomaly is effective in relieving symptoms of TOS.

Clavicular anomalies. Clavicular fractures are common injuries, yet only a few case studies have reported TOS as a secondary outcome to clavicular fracture malunion and ossification.[10,11] In these reports, magnetic resonance imaging reveals irritation of the brachial plexus and compression of the costoclavicular space due to bone callus formation and/or a protruding fragment of the clavicle.[10,11] Treatment plans varied between case reports, and no case recommended a consistent treatment plan for secondary thoracic outlet clavicular malunion.

In a retrospective review of 400 patients diagnosed with TOS, Weber and Criado[12] reported that 6.25% of those patients also possessed a clavicular anomaly, which is comparable to our findings. In addition, in the clavicular anomaly cohort described by Weber and Criado,[12] all patients had excision of the clavicular anomaly, yet only six had additional resection of the first rib. None of our patients underwent additional resection of the clavicular anomaly as we have found that the resection of the clavicle causes deformity and shoulder destabilization. After FRAAS in the clavicular cohort, all patients reported symptom resolution.

Cervical ribs. Less than 1% of the population has a cervical rib, and only 10% of those with cervical ribs are symptomatic.[2] Patients with TOS are often found to have a higher prevalence of accessory cervical ribs (range, 8.5%-29.5%) compared with the general population (0.74%).[4,5] The presence of a cervical rib increases the likelihood of experiencing TOS symptoms, but the cervical rib should only be resected if the patient is symptomatic on the side of the cervical rib. As seen in our patient with ATOS with bilateral cervical ribs and a fused right first and second rib, resection of the left cervical rib was not necessary as no TOS symptoms were present on that side.

According to a study by Chang et al,[13] different variants of TOS have been shown to coexist especially in patients with large cervical ribs. As noted in the study, 20 of 423 patients with TOS were identified with a cervical rib, and all obtained surgical removal of the cervical and first rib with great outcomes.[13] Furthermore, all subsets of TOS were represented, and all patients with ATOS had concomitant neurogenic symptoms of pain and weakness.[13] Likes et al[14] reported that a small subset of patients had an arterial component in addition to their neurogenic symptoms. As also seen in our retrospective review, the coexistence of different TOS variants is not uncommon in patients with boney abnormalities and treatment should be tailored accordingly.

Residual rib. The presence of a residual first rib after TOS operative management creates an increased risk for reoperation. In fact, the most common cause of TOS recurrence is a residual first rib.[15] Urschel and Razzuk[15] discovered that 87% of 1221 TOS recurrences were due to a residual first rib. Similar results were observed by Mingoli et al,[16] in which 80% of patients identified with a residual first rib via chest radiography had recurrent symptoms.

The high recurrence of symptoms from a residual rib is explained by the anatomy of the disease. As observed in our review, a residual first rib fragment leads to continued swelling, pain, and discomfort due to the ability of scar tissue to anchor onto the rib.[17] In order to alleviate the recurrent symptoms, the removal of the residual rib and surrounding fibrous scar tissue is required. In the 87% of TOS recurrences treated by Urschel and Razzuk,[15] 97.1% of those patients experienced symptom resolution after the resection of the residual first rib. Similarly, in Mingoli et al,[16] 100% of the

patients with recurrent TOS symptoms due to a residual rib experienced symptom improvement after resection. Our results support both studies as our patient with a residual rib had complete symptom resolution after surgery. This portrays the importance of complete excision of the first rib and scalene muscles within the first operation to avoid the continued compression of the interscalene space and poor surgical outcomes.

CONCLUSIONS

Patients with boney abnormalities can present with any of the three forms of TOS: neurogenic, arterial, and venous. Some patients will have more than one of these presentations concomitantly. As evidenced by our study, it appears that the appropriate removal of the boney abnormality provides excellent results, making it an essential intervention for patients with concomitant forms of TOS.

REFERENCES

1. Jones MR, Prabhakar A, Viswanath O, Uritis I, Green JB, Kendrick JB, et al. Thoracic outlet syndrome: a comprehensive review of pathophysiology, diagnosis, and treatment. Pain Ther 2019;8:5-18.
2. Hooper TL, Denton J, McGalliard MK, Brismée JM, Sizer PS Jr. Thoracic outlet syndrome: a controversial clinical condition. Part 1: anatomy, and clinical examination/diagnosis. J Man Manip Ther 2010;18:74-83.
3. Sanders RJ, Hammond SL, Rao NM. Diagnosis of thoracic outlet syndrome. J Vasc Surg 2007;46:601-4.
4. Brewin J, Hill M, Ellis H. The prevalence of cervical ribs in a London population. Clin Anat 2009;22:331-6.
5. Makhoul RG, Machleder HI. Developmental anomalies at the thoracic outlet: an analysis of 200 consecutive cases. J Vasc Surg 1992;16:534-42; discussion: 542-545.
6. Atasoy E. Recurrent thoracic outlet syndrome. Hand Clin 2004;20:99-105.
7. Terabayashi N, Ohno T, Nishimoto Y, Oshima K, Takigami L, Yasufku Y, et al. Nonunion of a first rib fracture causing thoracic outlet syndrome in a basketball player: a case report. J Shoulder Elbow Surg 2010;19:e20-3.
8. Mirza AK, Duncan AA. Thoracic outlet syndrome as a consequence of isolated atraumatic first rib fracture. J Surg Case Rep 2017;2017:rjx100.
9. Hines K, Graf E, Liu D, Freischlag JA. The rare case of cervical rib fusion to the second rib. Ann Vasc Surg 2014;28:742.e5-8.
10. Beliaev AM, Fougere C. Thoracic outlet syndrome secondary to a mid-clavicle malunion. BMJ Case Rep 2015;2015. bcr2015209583.
11. Yoo MJ, Seo JB, Kim JP, Lee JH. Surgical treatment of thoracic outlet syndrome secondary to clavicular malunion. Clin Orthop Surg 2009;1:54-7.
12. Weber AE, Criado E. Relevance of bone anomalies in patients with thoracic outlet syndrome. Ann Vasc Surg 2014;28:924-32.
13. Chang KZ, Likes K, Davis K, Demos J, Freischlag JA. The significance of cervical ribs in thoracic outlet syndrome. J Vasc Surg 2013;57:771-5.
14. Likes K, Rochlin DH, Call D, Freischlag JA. Coexistence of arterial compression in patients with neurogenic thoracic outlet syndrome. JAMA Surg 2014;149:1240-3.
15. Urschel HC Jr, Razzuk MA. The failed operation for thoracic outlet syndrome: the difficulty of diagnosis and management. Ann Thorac Surg 1986;42:523-8.
16. Mingoli A, Feldhaus RJ, Farina C, Cavallari N, Sapienza P, Di Marzo L, et al. Long-term outcome after transaxillary approach for thoracic outlet syndrome. Surgery 1995;118:840-4.
17. Likes K, Dapash T, Rochlin DH, Freischlag JA. Remaining or residual first ribs are the cause of recurrent thoracic outlet syndrome. Ann Vasc Surg 2014;28:939-45.

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APPENDIX

Supplementary Table I (online only). Boney abnormalities present within the thoracic outlet syndrome (*TOS*) database 2016-2021

TOS patients	No. (%) (N = 73)
Residual rib	1 (1.37)
Fractured rib	3 (4.11)
Cervical rib	3 (4.11)
Clavicular fracture	4 (5.48)
Nonfused congenital clavicle	1 (1.37)
No boney abnormality	61 (83.56)

Supplementary Table II (online only). Demographic characteristics of patients with thoracic outlet syndrome (*TOS*) in the boney abnormality cohort

TOS presentation (N = 12)	ATOS (n = 1)	VTOS (n = 5)	NTOS (n = 6)
Sex, No. (%)			
Male	–	3 (60)	3 (50)
Female	1 (100)	2 (40)	8 (50)
Median age, years (range)	38	30 (16-37)	18 (14-60)
Median symptom duration, months (range)	4	5 (0.16-144)	8 (5-12)
Boney abnormality, No. (%)			
Residual first rib	–	1 (20)	–
Fractured first rib	–	–	3 (50)
Fused ribs	1 (100)[a]	–	–
Cervical ribs			2 (33)
Unilateral	–	–	–
Bilateral	1 (100)[a]	–	
Clavicular anomaly	–	1 (20)	–
Congenital clavicle	–	3 (60)	1 (17)
Clavicular fracture	–	–	–
Boney abnormality etiology, No. (%)			
Congenital	1 (100)	1 (20)	2 (33)
Traumatic/surgical	–	4 (80)	4 (67)
Surgical approach, No. (%)			
Transaxillary	1 (100)	4 (80)	6 (100)
Infraclavicular	–	1 (20)	–
Complications, No. (%)			
Pneumothorax	1 (100)	3 (60)	3 (50)
Infection	–	1 (20)	–
Length of stay, days (range)	2	1 (1-2)	1 (1-2)
Length of anticoagulation, months (range)	6.6	4 (1.6-6.2)	N/A
Length of follow-up, months (range)	6.6	6 (5-25)	1 (0.4-8)

ATOS, Arterial thoracic outlet syndrome; *N/A*, not applicable; *NTOS*, neurogenic thoracic outlet syndrome; *VTOS*, venous thoracic outlet syndrome.
[a]This patient with ATOS presented with a fused first and second rib as well as cervical ribs.