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An unusual case of neurogenic thoracic outlet syndrome



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ABSTRACT

INTRODUCTION: Neurogenic thoracic outlet syndrome (nTOS) is the most common manifestation of thoracic outlet syndrome (TOS), accounting for more than 95% of cases. It is usually caused by cervical ribs, anomalies in the scalene muscle anatomy or post-traumatic inflammatory changes causing compression of the brachial plexus.

CASE PRESENTATION: We present an unusual case of nTOS caused by a cystic lymphangioma at the thoracic outlet, with only one case reported previously in the literature. We used a combined supraclavicular and transaxillary approach for complete removal, which resulted in excellent recovery of the patient.

DISCUSSION: Though lymphatic cysts may be commonly encountered in surgical practice, compression causing nTOS is extremely rare. The location of the lymphatic cyst with compression of the brachial plexus may provide a challenge for treatment. Surgical excision is the preferred method of management, with higher success rates than sclerotherapy.

CONCLUSION: Surgical excision to ensure complete removal of the cyst is recommended. Sclerotherapy may be used in cases where complete excision of the cyst wall may not be possible.

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1. Introduction

Thoracic outlet syndrome (TOS) is caused by compression of neurovascular structures in the region of the thoracic outlet. Depending on the pathology at the thoracic outlet, there were several terms coined such as cervical rib syndrome [1], scalenus anticus (Naffziger) syndrome [2], hyperabduction syndrome [3] and costoclavicular syndrome [4]. Neurogenic thoracic outlet syndrome (nTOS) is a peripheral compressive neuropathy of the brachial plexus at the thoracic outlet. It results from congenital cervical ribs, anatomical anomalies of the scalene muscles or inflammation caused from trauma leading to compression of the brachial plexus [5]. A lymphatic cyst causing nTOS is an extremely rare condition, with only one case reported previously [6]. We present this case report to highlight the importance of complete excision of the cyst wall without injuring the neurovascular structures, and describe the combined supraclavicular and transaxillary approach for excision. This work has been written in accordance with the SCARE criteria [7].

2. Case presentation

A 24-year-old, otherwise healthy female presented to our hospital with complaints of gradually progressive tingling and numbness over the fifth finger of her left hand and medial aspect of the wrist for three months. These symptoms worsened on performing daily activities using her left upper limb. She had noticed a vague fullness over her left supraclavicular area since the onset of her symptoms. There was no history of trauma or other comorbidities.

On examination, there was a vague swelling over the left supraclavicular region. There were no signs of sensory or motor deficit in the left upper limb. There was no evidence of muscle wasting, temperature changes or edema. However, Adson's test showed absence of the left radial pulse. A neck radiograph showed no cervical rib. Arterial duplex scan showed mild compression of the left subclavian artery, but no evidence of thrombosis. MRI of the neck showed a cystic swelling at the left thoracic outlet causing compression of the brachial plexus nerve trunks, with extension into the left axilla, with features suggestive of a lymphatic cyst (Figs. 1 and 2). There was no evidence of arterial compression on the MRI.

She underwent excision of the lymphatic cyst. A supraclavicular approach was used to dissect the cyst away from the neurovascular structures in the neck. The trunks of the brachial plexus were found to be lifted and splayed by the tense cyst, posing difficulty in dissection of the cyst wall. Hence a combined trans-axillary approach was carried out to mobilize the cyst adequately. The procedure was completed successfully without any injury to the brachial plexus, with complete removal of the cyst. The postoperative course was

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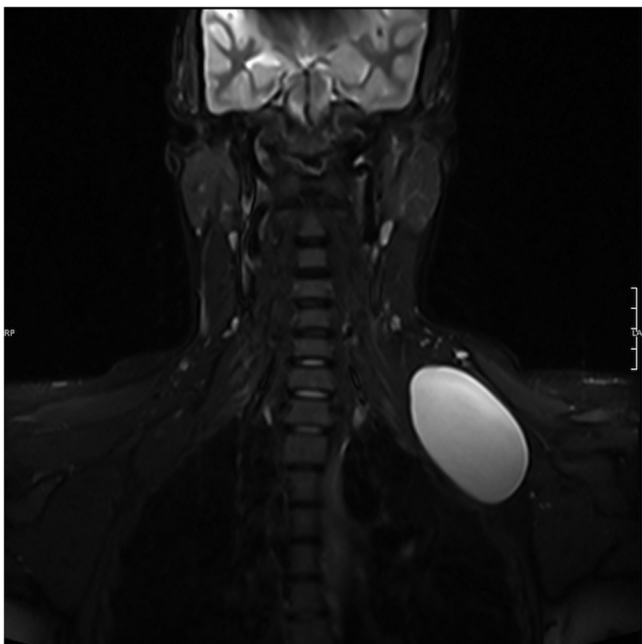


Fig. 1. Lymphatic cyst compressing brachial plexus and extending into axilla.

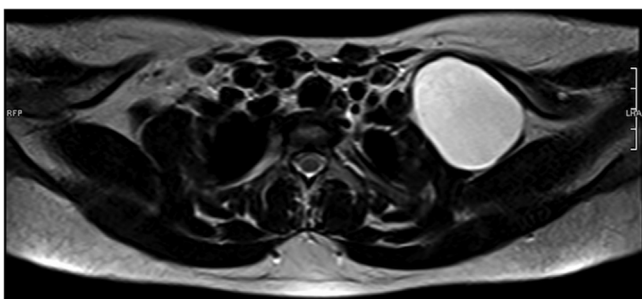


Fig. 2. Axial section image of lymphatic cyst.

uneventful. Histopathology showed benign lymphoid cells, suggestive of lymphatic cyst.

3. Discussion

Thoracic outlet syndrome (TOS) is a collection of symptoms and signs that result from compression of neurovascular structures at the thoracic outlet. Neurogenic thoracic outlet syndrome is the most common presentation, occurring in more than 95% cases of TOS. It results from compression of the brachial plexus. Vascular TOS, either venous or arterial account for the remainder, forming 3% and 1% of TOS cases respectively [5]. A large study involving 200 patients who underwent surgery for TOS found that the majority of cases (43%) were due to variations in scalene and subclavius muscle insertions. 10% cases had supernumerary scalene muscles, and 8.5% cases were attributed to cervical rib [8].

Acquired causes of TOS include post-traumatic inflammation most commonly following hyperextension injury of the neck [9]. Lymphocele causing nTOS is an extremely rare condition, with only a single case report published so far in the literature [6].

Lymphoceles, also termed cystic lymphangiomas, are congenital malformations occurring from lymph accumulation in the jugular lymph sacs, due to obstruction of the fetal lymphatics. These primitive lymph sacs do not connect with the venous system, resulting in development of lymphocele. They are very rare cystic lesions in adults, with most seen during childhood.

Surgical excision is routinely done for lymphatic cyst in the neck to prevent further growth or recurrence [10]. Sclerotherapy has also been tried with varying results. Karcaaltincaba et al. recommended using ethanol, tetracycline, talc or fibrin glue for pelvic lymphoceles [11]. Ogita et al. introduced the immunostimulant OK-432 (Picibanil) as a sclerosing agent for these lesions [12]. They found complete regression in 8 of the 9 patients treated after 2–3 months. A recent study of 131 patients with lymphatic malformations treated with OK-432 showed a successful outcome only in 70% of the patients [13].

Hence, surgery is considered the preferred treatment option. Various approaches to the thoracic outlet have been described including supraclavicular, transaxillary and infraclavicular [14]. Orlando et al. reported results of transaxillary surgical intervention in 538 patients of TOS with 594 first rib resections done. 308 (52%) patients had neurogenic TOS and excellent results were observed, with improved or fully resolved symptoms in 93–96% cases [15].

However, Qvarfordt et al. found different results. 97 patients underwent transaxillary approach alone for neurogenic TOS, but a fifth had persistent or recurrent symptoms necessitating reoperation by the supraclavicular approach. They subsequently combined the supraclavicular and transaxillary approach in 94 patients with excellent results [16]. We combined the former two techniques to separate the cyst wall from the neurovascular structures in the neck, thus allowing complete excision of the cyst.

4. Conclusion

We would like to highlight the fact that lymphatic cyst in neck may produce nTOS and should be kept in the differential diagnosis. The recommended treatment is complete surgical excision of the cyst to avoid recurrence. Sclerotherapy may be considered as an alternative if complete excision becomes technically challenging.

Conflict of interest

There are no conflicts of interest to report.

Funding

None.

Consent

Informed consent was obtained from the patient for presentation of the details of this case, along with the images for the purposes of publication. No personal identification information has been displayed in the images.

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Author contribution

Both the authors namely, Dr. Yash Vaidya and Dr. Rajan Vaithianathan, were involved in the management of this patient. This manuscript has been drafted by both authors.

Ethical approval

Ethical approval obtained.

Registration of research studies

Not applicable.

Guarantor

Dr. Yash Vaidya.

References

- [1] J.B. Murphy, Cervical rib excision: collective review on surgery of cervical rib, *Clin. John B Murphy* 5 (1916) 227–240.
- [2] A. Ochsner, M. Gage, M. DeBakey, Scalenus anticus (Naffziger) syndrome, *Am. J. Surg.* 28 (June) (1935) 669–695.
- [3] M.A. Falconer, G. Weddell, Costoclavicular compression of the subclavian artery and vein, *Lancet* 2 (October) (1943) 539–544.
- [4] I.S. Wright, The neurovascular syndrome produced by hyperabduction of the arms. The immediate changes produced in 150 normal controls, and the effects on same persons of prolonged hyperabduction of the arms, as in sleeping, and in certain occupations, *Am. Heart J.* 29 (January) (1945) 1–19.
- [5] R.J. Sanders, S.L. Hammond, N.M. Rao, Diagnosis of thoracic outlet syndrome, *J. Vasc. Surg.* 46 (September (3)) (2007) 601–604.
- [6] M.B. Marshall, T. Oliveira, Thoracic outlet lymphangioma, *Interact Cardiovasc. Thorac. Surg.* 9 (August (2)) (2009) 372–373.
- [7] R.A. Agha, A.J. Fowler, A. Saeta, I. Barai, S. Rajmohan, D.P. Orgill, SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (October) (2016) 180–186.
- [8] R.G. Makhoul, H.I. Machleder, Developmental anomalies at the thoracic outlet: an analysis of 200 consecutive cases, *J. Vasc. Surg.* 16 (October (4)) (1992) 534–542.
- [9] L. Casbas, X. Chauffour, J. Cau, J.P. Bossavy, D. Midy, J.C. Baste, A. Barret, Post-traumatic thoracic outlet syndromes, *Ann. Vasc. Surg.* 19 (January (1)) (2005) 25–28.
- [10] A. Makni, F. Chebbi, F. Fetirich, R. Ksantini, H. Bediou, M. Jouini, M. Kacem, Z. Ben Safta, Surgical management of intra-abdominal cystic lymphangioma. Report of 20 cases, *World J. Surg.* 36 (May (5)) (2012) 1037–1043.
- [11] M. Karcaaltincaba, O. Akhan, Radiologic imaging and percutaneous treatment of pelvic lymphocele, *Eur. J. Radiol.* 55 (September (3)) (2005) 340–354.
- [12] S. Ogita, T. Tsuto, K. Tokiwa, T. Takahashi, Intracystic injection of OK-432: a new sclerosing therapy for cystic hygroma in children, *Br. J. Surg.* 74 (August (8)) (1987) 690–691.
- [13] N. Ghaffarpour, B. Petrini, L.A. Svensson, K. Boman, T. Wester, G. Claesson, Patients with lymphatic malformations who receive the immunostimulant OK-432 experience excellent long-term outcomes, *Acta Paediatr.* 104 (November (11)) (2015) 1169–1173.
- [14] R.J. Sanders, W.H. Pearce, The treatment of thoracic outlet syndrome: a comparison of different operations, *J. Vasc. Surg.* 10 (December (6)) (1989) 626–634.
- [15] M.S. Orlando, K.C. Likes, S. Mirza, Y. Cao, A. Cohen, Y.W. Lum, T. Reifsnnyder, J.A. Freischlag, A decade of excellent outcomes after surgical intervention in 538 patients with thoracic outlet syndrome, *J. Am. Coll. Surg.* 220 (May (5)) (2015) 934–939.
- [16] P.G. Qvarfordt, W.K. Ehrenfeld, R.J. Stoney, Supraclavicular radical scalenectomy and transaxillary first rib resection for the thoracic outlet syndrome. A combined approach, *Am. J. Surg.* 148 (July (1)) (1984) 111–116.

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