Neurogenic thoracic outlet syndrome: Are anatomical anomalies significant?

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Background. Thoracic outlet syndrome (TOS) is one of the most poorly understood syndromes. Neurogenic TOS is found in 95% of cases. The described anatomical spaces transform and evolve into 'entrapment spaces'. The aetiology is unclear. This study was based on the observation by a single surgeon that there appeared to be a high incidence of anatomical abnormalities in patients with neurogenic TOS.
Objective. To attempt to clearly define anatomical anomalies causing TOS.
Methods. The records from a prospectively maintained computer database of 219 patients submitted for surgery over a 10-year period (1999 - 2009) were reviewed. A substudy was done on the patients operated on over the last 4 years (n=63) in whom details of the intraoperative anatomical findings were meticulously recorded.
Results. Over the last 4 years, the surgical findings in the last 63 patients (67 operations) revealed a significant number of anatomical abnormalities believed to be responsible for the nerve compression. Brachial plexus anomalies were found in 99% of the patients – the majority comprised the postfixed configuration. In addition, 58% had a soft-tissue anomaly, 27% had a bony anomaly and 3% had other abnormalities. The majority had combinations of these abnormal findings.
Conclusion. These findings strongly suggest that there is usually an identifiable anatomical cause, typically the brachial plexus, for the symptoms of TOS. We strongly recommend that the supraclavicular approach be used in order to define anatomical aberrations. Brachial plexus configuration anomalies causing TOS have not been emphasised previously. Further detailed recordings of these findings may help us better understand the aetiology of this poorly defined syndrome.

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Thoracic outlet syndrome (TOS) is controversial in terms of definition, anatomy, aetiology and treatment. The definition of TOS is generally accepted as 'upper extremity symptoms due to compression of the neurovascular bundle in the area of the neck just above the first rib'.[1] The thoracic outlet, also known as the cervicothoracobrachial junction, consists of three important compartments through which vital structures such as nerves and blood vessels run. These compartments are the interscalene space, the costoclavicular space and the retropectoralis minor space. Neurogenic TOS, resulting from nerve compression, is the most common pathology, accounting for >95% of TOS cases.[1] The majority of cases result from anatomical distortion at the interscalene triangle.[2] For various reasons, the described anatomical spaces transform and evolve into 'entrapment spaces'. The aetiology of this is largely unclear, but it is thought that changes may be congenital or acquired, and that these alterations involve either the bony structures or the soft tissues.[1,2] The observation of anatomical anomalies, especially of the brachial plexus, initiated this study.

The aetiology of neurogenic TOS is multifactorial, with bony tissue abnormalities and soft-tissue abnormalities described as definite contributors to the syndrome. These abnormalities contribute to the syndrome by altering the space within which the brachial plexus trunks run. Brachial plexus anomalies, however, have not classically been described in direct association with TOS. Recent interest in brachial plexus anomalies suggests them as a cause for various pathologies, including TOS.[3]

We hypothesised that brachial plexus anomalies, alone or in conjunction with additional pathology, are almost always associated with neurogenic TOS. The objective of this study was to attempt to clearly define anatomical anomalies causing TOS.

Methods

The study design is a retrospective review from a prospectively maintained computer database of patients presenting with TOS, over a 10-year period. From the computer database, the records of all patients with TOS were scrutinised, and information regarding their preoperative symptoms, investigations, operative surgery and clinical outcomes documented.

The neurological presentation of pain, paraesthesia, weakness, wasting of the hand muscles and the segmental distribution in conjunction with the clinical investigations (including the elevated arm stress test), investigations (a chest radiograph, magnetic

resonance imaging and nerve conduction studies) and the pathology (cervical rib, fused rib) have been correlated with the surgical procedures of neurolysis with cervical or first rib resection.

All data were captured into an Excel (Microsoft, USA) spreadsheet and subjected to multivariate analysis.

A total of 219 patients were analysed from the computer database of a single vascular surgical practice. A subset of 63 patients, treated over the latter 4 years of the study, had a detailed assessment and evaluation of the anomalies seen at surgery. This is reported separately.

All operations were performed using a supraclavicular approach, which allowed identification of the brachial plexus and soft-tissue anomalies. All patients underwent anterior scalenectomy. If a cervical rib was present, it was resected. A first rib resection was performed in the majority of the patients; only three patients had division of fibrous bands in isolation. All patients had neurolysis performed. Each nerve root from C5 to T1 was adequately dissected out and mobilised to avoid persistent symptoms. No cervical sympathectomies were performed – this is a selective procedure reserved for patients with autonomic disturbances.

Ethics approval was obtained from the Biomedical Research Ethics Committee of the University of KwaZulu-Natal.

Results

There was a total of 219 patients, of whom 160 (73%) were women. The ages ranged from 5 to 63 years (mean 31).

All but one patient had pain or paraesthesia as the presenting complaint, with pain being the dominant symptom.

The anatomical findings were scrutinised at surgery, and varying degrees of bony, soft-tissue and brachial plexus abnormalities were found in all but one patient. The patient who had no abnormality detected had fibrosis in the interscalene space, which seemed to be restricting nerve mobility. Scar tissue is known to cause compression and neurogenic irritation.[4]

Subgroup of 63 patients identifying anatomical abnormalities

A total of 17 patients (27%) had previous injury to the neck. Eleven patients had undergone previous surgery to the neck or shoulder region – mostly anterior spinal fusions. Six patients (10%) experienced previous trauma (Table 1). Trauma included a shoulder dislocation, motor vehicle accident (with presumed whiplash) and clavicle fracture.

All patients had chest radiographs performed, of which 14 (22%) were abnormal (Table 2).

Table 1. Predisposing injury

Previous injurious event	Total patients (*N*=63), *n* (%)*
Previous surgery (cervical spine/shoulder)	11 (17)
Previous trauma	6 (10)
Total	17 (27)

*Percentages to the nearest whole number.

Operative management

Sixty-seven operations were performed on the 63 patients; 4 patients had bilateral disease that was treated with staged operations. Forty-nine patients (73%) had 1st rib resections and 14 (21%) had cervical rib resections. One patient had a combined

Table 2. Abnormalities seen on CXR

CXR abnormalities	Total patients (*N*=63), *n* (%)*
Cervical ribs	11 (17)
Complete	4 (6)
Incomplete	1 (2)
Bilateral	6 (9)
High insertion of 1st rib	1 (2)
Residual stump of 1st rib	1 (2)
Mild spondylosis	1 (2)

CXR = chest X-ray.
*Percentages to the nearest whole number.

Table 3. Details of surgical procedure

Procedure	*n* (%)*
1st rib resection	49 (73)
Cervical rib resection	14 (21)
Division of bands only	3 (4)
Combined cervical and rib resection	1 (2)

*Percentages to the nearest whole number.

Table 4. Anatomical abnormalities identified at surgery (*N*=67)

Anatomical abnormality	*n* (%)*
Bony abnormality	17 (25)
Cervical rib	15 (22)
Fused 1st and 2nd rib	2 (3)
Soft-tissue abnormality	39 (58)
Bands	10 (15)
Abnormal muscle mass	29 (43)
Brachial plexus abnormality	66 (99)
Postfixed brachial plexus	41 (61)
Prefixed brachial plexus	14 (21)
Other brachial plexus abnormalities	11 (17)
Other abnormalities	2 (3)
Subclavian artery compressed lower trunk	1 (1.5)
Fibrosis alone	1 (1.5)
Total	124 (100)

*Percentages to the nearest whole number.

cervical and 1st rib excision (2%). Three patients (4%) had division of fibrous bands alone, without rib resection (Table 3).

Operative findings at surgery that were considered to be contributory to symptomatology (Table 4) include bony abnormalities, abnormal muscle configurations and brachial plexus abnormalities. The majority of patients had combinations of these abnormal findings.

Seventeen patients had bony abnormalities, of which 15 were cervical ribs (complete or incomplete). Two patients had fused 1st and 2nd ribs, which formed a bony mass and confined the thoracic outlet space.

Over half of the patients were found to have a soft-tissue abnormality (58%). These included fibrous bands in 10 (15%) patients and abnormal muscle masses in 29 (43%) patients. The muscle abnormalities included hypertrophy of the scalenus muscle complex, amalgamation of the scalene muscles and interdigitation of the nerve roots through the muscle. This was found in addition to the other anatomical abnormalities.

For purposes of intraoperative labelling, a prefixed brachial plexus refers to an enlarged upper trunk that is fused with the middle trunk. A postfixed brachial plexus refers to an enlarged lower trunk that is fused with the middle trunk. In the dissection, the origin of the roots cannot be seen; it is assumed that these surgical descriptions of prefixed and postfixed brachial plexus may have higher or lower origins, respectively, compared with a normal brachial configuration.

Interestingly, all but one of the patients was considered to have some form of brachial plexus abnormality:

- The majority of these were postfixed brachial plexus (62%) (Fig. 1).
- The prefixed brachial plexus anomalies were far fewer (21%) (Fig. 2).
- Other anomalies included abnormal position of the brachial plexus within or around the scalene muscles, abnormal configurations of the roots, trunks or branches, and attenuated roots not fitting the description of a pre- or postfixed brachial plexus.

There was a single unusual anomaly where the subclavian artery was compressing the nerve trunk. One patient was found to have fibrosis alone and no other anatomical abnormalities; the fibrosis caused confinement of the thoracic outlet space.

Discussion

The focus of this review was on neurogenic TOS. The majority of patients were female, with an average age of 31 years, which mirrors the epidemiology in the literature. Neurogenic TOS is reported to occur four times more commonly in females, typically presenting between the ages of 20 and 40 years.[5]

Pain and paraesthesia are the main presenting symptoms of neurogenic TOS. A significant proportion of patients had either previous cervical spine or shoulder surgery or trauma (including whiplash, clavicular fractures and shoulder dislocation), which may have been factors contributing to their pathology. The predominant neurogenic distribution was C8/T1, which is in keeping with the literature, as this lower nerve trunk is the one susceptible to being stretched over the first rib.[2,5,6] In terms of treatment of these patients, surgery was the mainstay of management; patients had been referred through various disciplines, and conservative trials had already failed by this stage. Patients with TOS should be given a 3-month trial of conservative management before surgical intervention, unless the condition interferes with their daily quality of life (working or sleeping) or when they present with advanced signs (e.g. muscle wasting).[1]

The number of abnormal findings emphasises the importance of the supraclavicular approach, because a transaxillary incision, while a feasible therapeutic option, will not detect these abnormalities.[4]

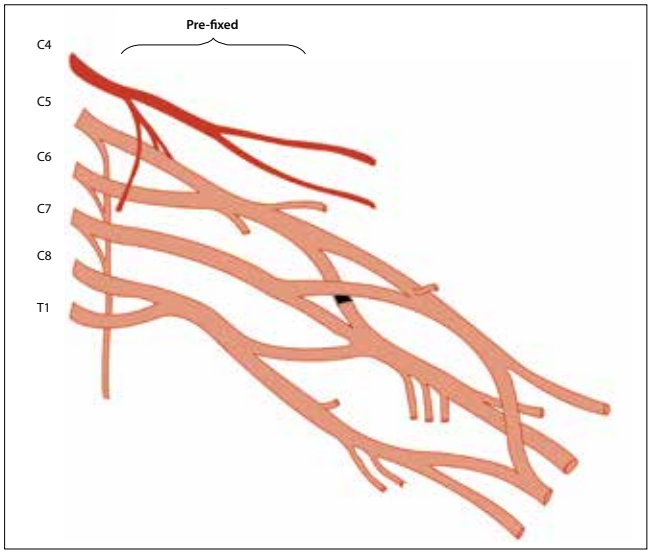

Fig. 1. Pre-fixed brachial plexus, indicating the contribution of C4 to the brachial plexus. (Adapted from Songcharoen P, Shin AY, Hand Surgery. 1st ed. [Online]. Philadelphia: Lippincott Williams & Wilkins, 2004. http://www.msdlatinamerica.com/ebooks/HandSurgery/sid744608.html#F3-57 (accessed 14 January 2015).)

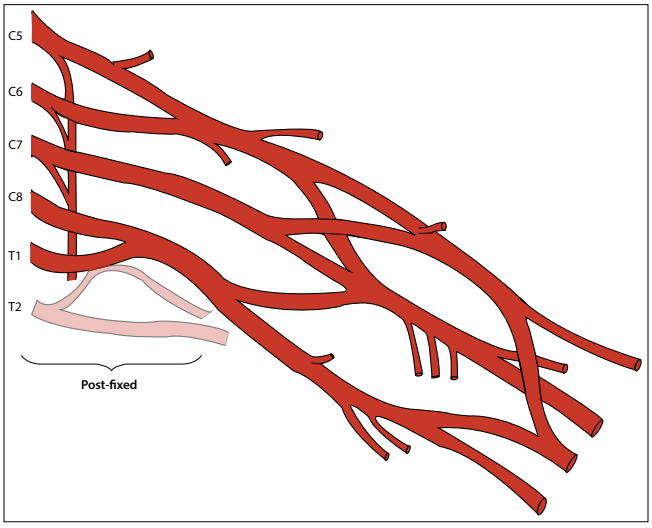

Fig. 2. Post-fixed brachial plexus, indicating the contribution of T2 from the brachial plexus. (Adapted from Songcharoen P, Shin AY, Hand Surgery. 1st ed. [Online]. Philadelphia: Lippincott Williams & Wilkins, 2004. http://www.msdlatinamerica.com/ebooks/HandSurgery/sid744608.html#F3-57 (accessed 14 January 2015).)

The findings at surgery included bony abnormalities, abnormal muscle configurations and brachial plexus anomalies. The majority of patients had combinations of these abnormal findings. It is thought that the outlet space, which becomes an 'entrapment space', may be from congenital or acquired causes. Most likely it is a combination of both factors: slight or grossly abnormal anatomy (congenital) coupled with hypertrophied muscles or a fibrotic process (acquired) that encases nerve structures with resultant pain.[1,2] Bony abnormalities have been well described as a cause or contributing factor in neurogenic TOS.[1,2] Cervical and fused 1st and 2nd ribs were both identified in this study. Soft-tissue abnormalities include scalene muscle abnormalities and fibrous bands. Such soft-tissue abnormalities have been documented in the literature, as well as the varying outlet dimensions between the anterior and medius scalene muscles.[1,2] Muscle abnormalities were found in the scalenus anterior, scalenus medius and sternocleidomastoid muscles.

Brachial plexus anomalies are not conclusively considered to be a factor contributing to neurogenic TOS; however, a recent article by Pellerin *et al*.[3] suggested that brachial plexus abnormalities are extremely common and that this anatomy may be a risk factor for certain pathologies, including TOS. The most common abnormality was a postfixed brachial plexus, implying a large conjoined middle and inferior trunk, susceptible to pathological stretching over the 1st rib or an anomalous cervical rib. Prefixed brachial plexus anomalies were also fairly common in patients presenting with symptoms attributable to the higher nerve roots, or in patients whose symptoms were not distinctively attributable to specific nerve roots within the brachial plexus. The remainder of brachial plexus anomalies described include combinations of nerve root formation that differ from pre- and postfixed brachial plexus configurations. Other brachial plexus abnormalities were of a minor nature and were related to the abnormal position of nerve roots (e.g. nerve roots arising anterior to the anterior scalenus muscle) or to the course of nerve roots (e.g. nerve roots interdigitating within the anterior scalenus muscle or scalenus medius muscle). The brachial plexus traversing the anterior scalenus muscle has been described, which was also noted in this study.[2]

Acquired abnormalities contributed to a large number (27%) of patients who had undergone either previous neck or shoulder surgery or who had trauma to the neck or shoulder region with resultant fibrosis, abnormal bone healing and muscle spasm. In addition, overuse caused hypertrophy of muscles in the interscalene space in a number of patients, confining the thoracic outlet space. These acquired abnormalities are the typical causes of TOS: post-traumatic scarring, postoperative scarring and repetitive trauma.[1,5] It is thought, however, that a predisposing factor is necessary for neck trauma to result in TOS. The predisposing factors described include bony and muscle abnormalities, both of which were noted in this study.[1]

Brachial plexus abnormalities fall under congenital anomalies, bony abnormalities may be congenital or acquired, but there is a rather indistinct line between congenital and acquired soft-tissue abnormalities. The majority of patients, even those with bony abnormalities, had more than one abnormal anatomical structure. Only two patients with cervical ribs had no other abnormalities seen at surgery. The patient who had no anatomical anomaly did have fibrosis, which was an explanation for the symptoms. This patient also responded well to surgery. In almost all patients, the classically described brachial plexus was not identified and this may well be a major factor contributing to neurogenic TOS.[3]

In keeping with the current literature, there were no deaths and complications were uncommon. Temporary neuropraxia was the most common complication.[2,4] Success rates with neurogenic TOS vary between 45 and 96% depending on the quoted series.[1,7] Success rates were excellent in this review with 96% of patients experiencing complete or partial resolution of symptoms.

Conclusion

We can conclude from this cohort of patients that there is always an underlying cause for the patients' symptoms, usually an anatomical abnormality. The majority of these anomalies are abnormalities of the brachial plexus. All patients with neurogenic TOS should therefore be considered as having 'true neurogenic TOS'. There is often more than one abnormality, even if subtle, making it likely that the pathology is multifactorial. For example, symptoms may develop in a person with an abnormal brachial plexus configuration that may become compressed later in life by muscle spasm or fibrosis after intense sport or a neck injury. In addition to this, the abnormality may only be detected by careful inspection of the anatomy at surgery. A supraclavicular approach is preferred, as a transaxillary approach precludes clear depiction of the neurovascular structures.

To clarify these findings, a further study is necessary to record detailed mapping of the brachial plexus findings intraoperatively. This then needs to be correlated with the specific presenting symptoms. Thirty-nine specific brachial plexus anomalies have been described in anatomy texts, emphasising that these anomalies may not be rare. The association between brachial plexus anomalies and neurogenic TOS symptoms can never be proven, as it is impossible to conduct a trial with a control group. However, the connection may be assumed if an anomaly is detected at surgery and the patient improves symptomatically thereafter.

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