Neurogenic Thoracic Outlet Syndrome caused by Subacute Clavicle Osteomyelitis

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Summary
A rare case of neurogenic thoracic outlet syndrome caused by subacute osteomyelitis (SOM) of clavicle is presented. It was treated by clavicle resection, 1st rib resection and scalenectomy followed by reconstruction of the clavicle by 6th rib vascularised bone transfer based on serratus anterior muscle as a rotational flap.

Key words: thoracic outlet syndrome, clavicle osteomyelitis, scalenectomy.

AIM OF THE DEMONSTRATION
According to Atasoy neurogenic thoracic outlet syndrome (TOS) is one of the most underrated, overlooked, and misdiagnosed conditions (1). It is estimated, that at least 0.1 % of the population are suffering from this condition(5). Unfortunately most clinicians are uncomfortable with the concept of the TOS. Not many physicians recognize this syndrome and even do not believe it exists, because frequently it is lacking any objective diagnostic criteria (8). It is considered to be a chameleon of syndromes, since it can imitate a number of pathologies, some of them very serious, thus correct differential diagnosis is of paramount importance (3). Most frequent cause of the TOS is trauma predisposed by soft tissue pathology which is followed by osseous pathology, such as cervical rib or elongated transverse process of C7 (9). Clavicle malunion and callus formation is a rare cause, but can produce compression of neurovascular structures with ease (6), (10).

Subacute osteomyelitis of the calvicle is a rare condition. To our knowledge there are no reports of TOS being caused by subacute osteomyelitis of the clavicle in the English literature.

The following TOS case caused by SOM of the clavicle was treated completely with the clavicle resection, 1st rib resection, anterior and middle scalenectomy and reconstruction of the clavicle with vascularised ipsilateral 6th rib.

CASE REPORT
20 year old female music college student was seen in the outpatient clinic complaining of nocturnal aching pain and tingling in the right arm for 7 years. Pain and paresthesia along the medial aspect of her right arm and ulnar digits was exacerbated after intensive piano lessons for many weeks. There was difficulty to abduct the shoulder due to a pain and progressive weakness. Full passive range of motion in the arm was possible. She was also complaining of deformed right clavicle due to a hematogenous osteomyelitis in the childhood. History also revealed needle biopsy of the lesion four years ago and curettage in other hospital three years ago that was followed with postoperative antibiotic treatment for 6 weeks. There was no other treatment since.

On physical examination bulging deformity of the right mid-clavicular area was noted. Some localized tenderness over the clavicle that was not associated with warmth, redness, and soft-tissue swelling. Tinel sign was positive on the right carpal, cubital tunnel and supraclaviculary above the anterior scalene muscle. Tinel on the pronator tunnel and infraclaviculary (on the pectoralis minor) was negative. Phalen sign was inconclusive, but Roos (EAST) provocation test was positive at 1 minute. There was a strong trigger point irritation on the right trapezius muscle. There were no pulse changes performing the Adson test. Two point discrimination was normal in all fingers. Electromyography (EMG) and nerve conduction study finding was nonspecific.

Plain radiographs in anteroposterior view showed bulging deformity of the mid-clavicle and typical radiographic appearance of a single, central transparent zone of osteolysis, well–marked outlines, and a zone of sclerotic bone representing subacute osteomyelitis (Fig.1.). Magnetic resonance imaging in coronal T1–weighted magnetic resonance images of the right brachial plexus showed signs of compression by the bulging portion of the mid–clavicle (Fig.2).

5 months of physical therapy, suspension of piano playing was ineffective. Patient considered her life to be miserable. She was ready to undergo major surgical procedure.

A diagnosis of neurogenic TOS secondary to subacute OM of the clavicle was made based on the patient’s history, physical examination, imaging studies. We planned to perform a clavicle resection, anterior and middle scalenectomy with transaxillary first rib resection, followed by clavicle reconstruction with vascularised VII rib transfer based on Serratus anterior muscle blood supply.

With the patient under general anesthesia, clavicle...
resection was performed. Wound was packed and draped with Opsite (Smith and Nephew) dressing. Next, the patient is placed with hips in the straight lateral position and the thorax tilted posteriorly 60 degrees with sandbag support at the back as described by Roos (7). The forearm, arm, axilla and chest are prepared and draped into the surgical field so it may be freely manipulated during operation. Incision was modified so that we could access to the axilla and harvest VII rib with 2 lower slips of Serratus anterior muscle. After the first rib resection, VII rib 12 cm long was harvested on the lateral thoracic artery. It was left in place and wound was packed with gauze dressing and covered with Opsite. The patient was placed supine again. Anterior and middle scalenectomy was performed via the supraclavicular approach. Both scalene muscles were sent for histological investigation. The tunnel was made between axilla and supraclavicular wound. Flap was passed through the tunnel and transferred rib was fixed to the remaining sternal and acromial part of the clavicle with plates and screws 2,4 mm (Fig.3 A and B).

During harvesting of VII rib pneumothorax was encountered. It was uneventfully treated with Pleuracan (B.Braun) system. Histology of the clavicle revealed subacute OM with mielofibrosis and atrophy with fibrosis in scalene muscles was diagnosed. Immediately after surgery patient was taken to intensive care unit for 3 days. Patient was discharged on 10th postoperative day with arm sling. 6 weeks after the surgery, pain and paresthesia in the right upper extremity had improved considerably and patient returned to piano practice. The patient is asymptomatic 9 months after surgery.

DISCUSSION
Claviculectomy for Thoracic outlet treatment has been described by Enker and Murthy (1970) (4). It is not practiced for treatment of the thoracic outlet syndrome nowadays, because resection of the clavicle will cause some degree of instability and disfigured aesthetic appearance, particularly in women. Today there are few accepted methods for decompressing “thoracic outlet area” – first rib resection (supraclavicular or infraclavicular) alone or in combination with scalenectomy. There is no uniform agreement on which procedure or combination is more effective, but some authors state that for upper type of Thoracic Outlet Syndrome scalenectomy alone will suffice. Others insist on doing complex approach to decompression of the area by removal of the first rib transaxillary and supraclavicular scalenectomy (2).

SOM is characterized by mild to moderate pain. Patient usually describes it as a persistent ache. Symptoms can be intermittent and onset is insidious. Often there is a long delay between the onset of pain and the diagnosis. Usually there are no constitutional symptoms. There may be many similarities between TOS and SOM of clavicle regarding pain: deep ache, night pain, insidious presentation and delay of diagnosis. In our case all symptoms were attributed to SOM and not TOS. Our patient was complaining of paresthesia and it is not a common symptom in SOM. Sometimes there are difficulties in differentiating subacut osteomyelitis from bone tumors such as osteoid osteoma, Ewing sarcoma or osteosarcoma, but in our case we were not having difficulty in diagnosis, since previous confirmatory biopsy have been performed. Magnetic resonance imaging was helpful not only in differentiating SOM from tumors, but also in localizing compression area (Fig.2.). It is not uncommon for EMNG to be negative in cases with confirmed direct intra–operative observation or preoperative MRI confirmation. For this reason EMNG should not be accepted a diagnostic tool. It has value only if it is positive.

In our case, an anterior and middle scalenectomy was performed including resection of the clavicle, first rib and reconstruction of clavicle with vascularised rib transfer. The patient obtained complete relief from her symptoms without major complications. Magnetic resonance imaging was not performed postoperatively to confirm straightening of distorted brachial plexus. There are authors who are in favor of scalenectomies alone in cases of posttraumatic TOS due to a callus formation at the clavicle fracture site(5). They are proposing that in clavicle fracture cases primary cause of TOS is scarring and contracture of scalene muscles and not a callus compression directly unto the underlying neurovascular structures in the costoclavicular space. In our case we were not able to resort to this advice since the clavicle itself has to be resected and reconstructed. At the same time scarred scalene muscles could not be left alone. To give even more space we decided to resect the 1st rib as well. It has been suggested by Atasoy that simultaneous transaxillary 1st rib resection and supraclavicular scalenectomy bears the best results in treatment of neurogenic TOS(6).

In this complex procedure full remission of symptoms were achieved by addressing both problems –TOS and SOM at the same time.

Conflict of interest: None

REFERENCES

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Fig. 1. Plain radiographs in anteroposterior view show bulging deformity of the mid–clavicle and typical radiographic appearance of a single, central transparent zone of osteolysis, well–marked outlines, and a zone of sclerotic bone representing subacute osteomyelitis

Fig. 2. Coronal T1–weighted magnetic resonance images of the right brachial plexus showed signs of compression (white arrow) by the bulging portion of the mid–clavicle

Fig. 3. A – supraclavicular area with visible 2,4 mm titanium fixation plate. Black arrow – Serratus anterior muscle supplying vascularised rib transfer. White arrow – preserved supraclavicular nerves. S – sternal end, A – acromial end of the reconstructed clavicle

Fig. 3. B – Plain radiographs in anteroposterior view show reconstructed clavicle with vascularised rib transfer and 2,4 mm titanium fixation devices