

Hypertrophic Non-union of the First Rib Causing Thoracic Outlet Syndrome

: A Case Report

We experienced a rare case of thoracic outlet syndrome caused by hypertrophic nonunion of the first rib. A diagnosis was made mainly upon provocative tests and imaging studies. Pain and tingling could be reproduced and the radial pulse obliterated by the hyperabduction test. Abundant callus formation on the posterior aspect of the first rib with fracture line was visible on plain radiograph. Two-dimensional computed tomography showed right thoracic outlet narrowing mainly caused by the mass-effect of the callus. Dynamic arteriographic studies revealed an external compression of the right subclavian artery and duplex ultrasonography demonstrated a reduction in right subclavian artery blood flow when the shoulder is in 90 degrees of abduction. Surgery was performed after the conservative management for three months which failed to relieve the patient of his complaints. Resection of the first rib via transaxillary approach was undergone uneventfully in combination with the myotomy of the scalenus anticus muscle. At postoperative one year follow up, the patient was free of symptoms, and had a full range of motion of the right shoulder with no evidence of arterial insufficiency.

Key Words : *Hyperostosis; Hypertrophic Nonunion; Ribs; Thoracic Outlet Syndrome*

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INTRODUCTION

Thoracic outlet syndrome (TOS) is a disorder referring to the clinical manifestations of the compression of great vessels and nerves at the base of the neck as they pass from the mediastinum and neck to the axilla. It was first described by Sir Astley Cooper in 1821 and numerous articles have been written regarding its causes, symptomatology, diagnosis, and treatment. Symptoms are generally mild but bothersome and diagnosis is sometimes difficult. A patient with clinical manifestations of intermittent neurovascular compromise resulting from a rare cause of TOS is described.

CASE REPORT

A 19-yr-old male slipped and fell during an altercation with his friend. He was apparently asymptomatic until eight months later when he started to complain of a tingling sensation on his right hand only while his shoulder was in an abducted position. Later he was dropping things from his right hand. After three months of medical treatment and physical therapy without improvement of the symptoms, he visited our institution.

Physical examination showed no limitation of motion of

the shoulder, elbow or hand. Neither deformities nor neuromuscular deficits were observed. Antero-posterior and lateral radiographs of the upper extremities showed an apparent fracture of the first rib with abundant callus formation and the fracture line still visible. On chest films, callus formation was seen on the posterior aspect of the first rib (Fig. 1A). Provocative tests for thoracic outlet syndrome like costoclavicular compression, neck tilting, and Adson's test were negative and only the hyperabduction test was positive. The radial pulse disappeared and the patient experienced pain and tingling in the right hand. Signs and symptoms were relieved when the shoulder was brought back to neutral.

Electromyography and nerve conduction studies failed to show any abnormalities. Two-dimensional computed tomography showed right thoracic outlet narrowing mainly caused by the mass-effect of the callus formation on the posterior aspect of the right first rib and the fracture line was likewise visible (Fig. 1A). Dynamic arteriographic studies revealed an external compression of the right subclavian artery on shoulder hyperabduction (Fig. 1B). Duplex ultrasonography demonstrated a reduction in right subclavian artery blood flow when the shoulder is in 90 degrees of abduction compared to when the shoulder is in neutral (Fig. 1C).

A diagnosis of TOS caused by the abundant callus formation on the posterior aspect of the first rib with the finding

of nonunion was made. Surgery was decided after the conservative management for 3 months which failed to relieve the patient of his complaints. A resection of the first rib and the myotomy of the scalenus anticus muscle on the right was contemplated to decompress the right thoracic outlet.

A transaxillary approach was utilized with the patient in a lateral position. The anterior origin of the serratus anterior muscle was detached from the second rib and the two intercostobrachial nerves were identified and preserved. The scalenus anticus muscle was cut by sharp dissection. The periosteum over the rib was incised and dissected. The anterior portion of the first rib was then osteotomized at the level of the scalene tubercle cautiously not to injure the subclavian vessels and brachial plexus. The posterior remainder of the first rib including the hypertrophied callus was removed piecemeal as much as possible with a rongeur. A chest radiography was taken to check any evidences of iatrogenic pneumothorax immediately after the surgery and showed no remarkable finding (Fig. 1D).

At postoperative one year follow up, the patient was free of symptoms, had a full range of motion of the right shoulder with no evidence of arterial insufficiency. No complication was seen.

DISCUSSION

The brachial neurovascular bundle runs under the clavicle and subclavian muscle (costoclavicular space) to the upper extremity after passing through the narrow interscalene triangle or thoracic outlet composed of the first rib in its inferior border, the scalenus medius muscle in its posterior border and the scalenus anticus muscle in its anterior border. If this space is narrowed or compressed intermittently or continuously for any reason there occurs a compromise of the brachial plexus, subclavian artery, or subclavian vein. Usual causative mechanism of TOS is congenital and trauma has only occasionally been implicated in producing TOS (1).

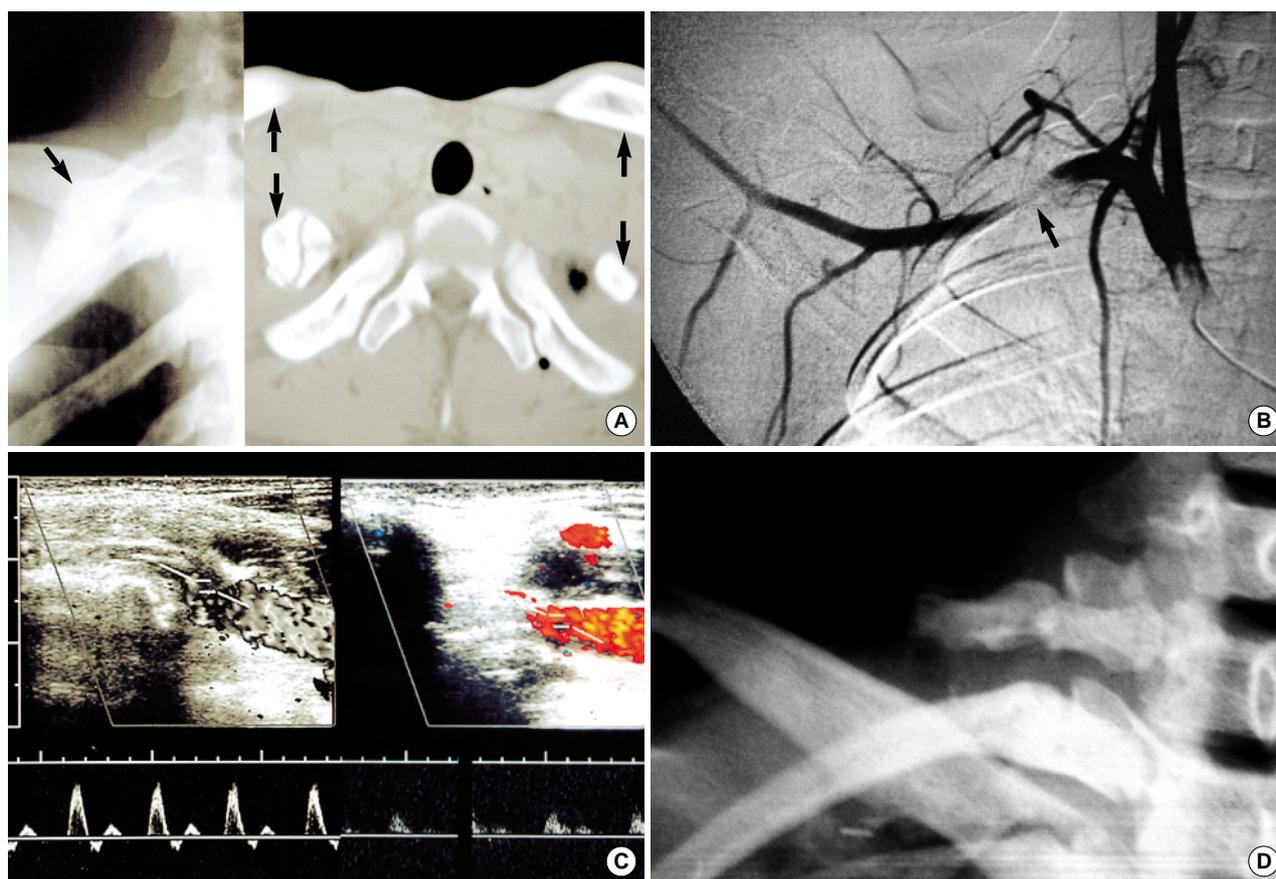


Fig. 1. (A) The plain radiograph shows hypertrophied callus on the posterior aspect of the first rib (black arrow). The distance between clavicle and first rib was 5 mm shorter than that of left side on computed tomogram (difference of the distance between the two arrows). Nonunion was confirmed by this computed tomographic film on which gap was marked between two fragments. (B) At this position of hyperabduction, arterial pulse of right radial artery disappears rapidly. Dynamic arteriography depicted disruption of the axillary arterial flow by its narrowing at this position as well (arrow). (C) The Duplex ultrasonography, which has a function of a color Doppler, demonstrates the compression of the subclavian artery as at least a doubling of peak systolic velocity and reduction of the flow at hyperabduction over 90 degrees. (D) Postoperative plain radiograph after resection of the first rib including the hypertrophied callus.

Traumatic disruption of the first rib potentially narrows the interscalene triangle, and may result in mechanical compression of the brachial plexus and subclavian artery. The reparative process with callus formation not only narrows this triangle but also may fix the subclavian artery. This tethering effect renders the artery more vulnerable to compression between the clavicle and the first rib with hyperabduction of the arm (1). Symptoms in the vast majority of TOS are mild, but they can be troublesome enough to make patients unable to work with their hands above their shoulders, to drive a car, or lift heavy objects. Sometimes the symptoms only manifest during provocations such as extreme ranges of shoulder and cervical motions which tend to stretch the neurovascular bundle and exaggerate the compression. Our patient also could not maintain his shoulder in the position of abduction over 90 degrees. Before attributing them to the compression of the neurovascular structures at the level of the thoracic outlet, differential diagnosis should include intrinsic neurologic, vascular, and musculoskeletal pathologies as well as the diseases caused by the compression of neural and vascular elements either proximal or distal to the thoracic outlet.

On physical examination, without provocative tests, findings might not be conclusive. Stallworth described that the Adson and Allen tests are not so helpful because they were positive in as many as 50% of normal population (2). Vascular compromise was demonstrated in our patient only during shoulder hyperabduction and there were no trophic changes in the distal extremity suggesting the lack of any longstanding static disorder. There was no atrophy in intrinsic muscles of hand, no hand weakness, and no clear sensory disturbances.

There is no reliable laboratory test for brachial plexus compression and no gold standard tool to diagnose TOS. Electromyography and nerve conduction studies are not known to be helpful in diagnosing TOS but are important in ruling out other causes. Likewise, electromyographic studies in our patient did not show findings of denervation and nerve conduction studies did not show slowing of conduction across the thoracic outlet. Plain radiograph may not be helpful, either. But, it can be valuable as a screening test particularly when the TOS is presented as a late developing sequelae with antecedent injury (1, 3).

Wood et al. emphasized the importance of additional evaluation to rule out other possible etiologies before an impression of TOS is made (4). Duplex ultrasonography is a diagnostic modality that demonstrates pulsating amplitude and frequency of a testing vessel at specific positions and functions as a color Doppler. Duplex ultrasonography would be able to show any turbulence and stenosis in the subclavian artery and is now considered to be increasingly important in the assessment of vascular symptoms due to subclavian compression (5). Color Doppler sonography is reported to be 92% sensitive and 95% specific for the diagnosis of the TOS

(6). Through these studies we were able to quantify the reduction of bloodflow and compare the differences of its value at different positions of the extremity. Angiogram is still regarded to be the most standard test for diagnosing TOS. On an angiogram, the patient with the arterial TOS usually reveals compressed area with poststenotic dilatation of the subclavian artery.

Once diagnosis has been made, surgery should be contemplated only after conservative management has failed. Moore pointed out that surgery should never be the first or even an early choice of treatment in his cases (7). According to the literature the data regarding physiotherapy is meager and whether it works is unclear. Thompson and Webster (5) suggested the first rib resection as an essential primary treatment for vascular complications of TOS. They reported that arterial involvement was not cured by the cervical rib excision alone and, whatever else was done, removal of the first rib was of vital importance (5). The first rib resection was also chosen for the posttraumatic pathology of the first rib (1, 3). Successful decompression could lead to an improvement in exercise tolerances of the elevated limb. However, prompt symptomatic improvement can not be expected post-operatively because of the chronicity of the compression.

Among several surgical approaches, the transaxillary approach is commonly used by surgeons because of its cosmetic merits and simplicity to get access to the first rib (1, 8). The advocates of the transaxillary approach sometimes suggest myotomy of the scalenus anticus muscle during the same operative sitting (9). We decided to perform the first rib resection through the transaxillary approach to decompress a narrowed thoracic outlet for the better access to the callus at the posterior portion of the first rib. Therefore the first rib was resected and the callus was removed in combination with the myotomy of the scalenus anticus muscle. The outcome of the patient was satisfactory with the gradual but complete relief of the neurovascular symptoms over a period of 5 months after the surgery.

In the diagnosis of TOS, exclusion of other disease entities should be made through various diagnostic modalities. Surgical removal of the cause of the compression should be done only after conservative therapies fail. Resection of the first rib via transaxillary approach in combination with the myotomy of the scalenus anticus muscle provided a satisfactory result in a case of TOS with hypertrophic nonunion of the first rib.

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