

“Clavicular Duplication Causing Thoracic Outlet Obstruction”: Unique Presentation of Unreported Association between Clavicular Duplication and Thoracic Outlet Syndrome

Agarwal S, Akhtar MN¹

Department of Orthopaedics and Trauma, North Eastern Indira Gandhi Regional Institute of Health and Medical Sciences, Shillong, Meghalaya, ¹Department of Orthopaedics, ESI Post Graduate Medical Institute of Health and Research, Kolkata, West Bengal, India

Address for correspondence:

Dr. Sharat Agarwal,
Department of Orthopaedics and
Trauma, North Eastern Indira
Gandhi Regional Institute of
Health and Medical Sciences,
Shillong - 793 018, Meghalaya, India.
E-mail: drsharat88@yahoo.com

Abstract

There are only few reports of clavicular duplication in the literature and all authors have agreed that duplicated clavicles are clinically insignificant. This is the first report of this unique association. A 22-year-old female student reported with features of neurogenic thoracic outlet syndrome mainly involving C8-T1 components of the brachial plexus, seemingly originating from involvement in costo-clavicular space. Radiograph of the shoulder revealed clavicular duplication. Neuro-physiological studies corroborated the diagnosis. Patient responded well to conservative treatment and was in follow-up for 1 year. Radiograph of the shoulder should be included in the workup of neurogenic thoracic outlet syndrome and anomalies of clavicle should be looked into and ruled out along with other common causes. Patient and symptom directed approach with dedicated physiotherapy and occupational therapy is required for optimum outcome.

Keywords: Bifid clavicle, Clavicular duplication, Neurogenic thoracic outlet syndrome, Thoracic outlet syndrome

Introduction

The clavicle develops from a cartilaginous anlage. It is the first bone to ossify from two primary centers, medial and lateral and a secondary center for sternal end. An osseous cuff develops very early in the middle part of the clavicle by the ossification in the perichondrium.^[1] Possibility of persistence of cartilaginous nest in the middle part of the clavicle has been hypothesized. There are only few reports of clavicular duplication in the literature with described etiology ranging from congenital, developmental and post traumatic, but all authors agreed on its clinical insignificance and mainly regarded it to be of incidental finding. During the diagnostic

work-up of neurogenic thoracic outlet syndrome, clavicular duplication was diagnosed. This case is presented to highlight the association between these two phenomena.

Case Report

A 22-year-old female student presented to orthopedic outpatient department (OPD) with the complaints of gradual onset neck and left shoulder pain radiating to hand, and associated with paresthesia and intermittent numbness of whole forearm and hand, but was worse around the ulnar border. For around 1 month, she noticed intermittent weakness and heaviness of limb associated with clumsiness in the left hand especially after overhead activities. Initially, the symptoms of radiating pain, paresthesia and numbness were less bothersome to the patient, but for 2 weeks it started increasing in intensity, so that patient attended OPD. There was no history of trauma to neck, shoulder or left upper limb. Patient neither had any history of headache, nocturnal awakening due to pain, cold intolerance, Raynauds phenomena, cyanosis, edema or claudication of the limb. Rounded contour of the shoulder was maintained with

Access this article online

Quick Response Code:



Website: www.amhsr.org

DOI:
10.4103/2141-9248.141980

some wasting of shoulder muscles. No supraclavicular fullness was present [Figure 1]. Wasting of hypothenar eminence was noted. Left hand was paler in color as compared to the right hand [Figure 2].

There was no neurological deficit but for decreased motor power (grade 2/5 on Medical Research Council scale) in 4th palmar interossei muscles (T1 root of brachial plexus), (card test) and decreased vibration sense in the left ring and little fingers (C8 root of brachial plexus) with intact proprioception. Thumb pressure for 30 s in the supraclavicular fossa in the supracoracoid region elicited radiating paresthesia in the forearm and hand, mainly of the ring and little fingers. She was not able to complete Roos test due to radiating paresthesia in the same region. Upper limb tension test and costoclavicular maneuver elicited the radiating paresthesia in the ring and little fingers, too. Adson's, Wright's, Halstead and Cyarix release tests were negative. Further clinical examination did not reveal glenohumeral, subacromial or cervical disc pathology. Peripheral nerve provocation tests for radial, median, and ulnar nerves were negative. Clinical diagnosis of neurogenic thoracic outlet syndrome was made. Cervical spine radiographs were normal. Anteroposterior radiograph of the left shoulder revealed clavicular duplication [Figure 3]. The nerve conduction study showed prolonged F-wave latency in the left ulnar and median motor nerves but normal compound muscle action potential amplitude. Decreased medial antebrachial cutaneous nerve (MABCN) sensory neural action potential amplitude with prolonged MABCN sensory latency (2.6 ms) was noted on MABCN conduction study. Needle electromyography showed abnormal spontaneous activities and long-duration motor unit potentials in the left first dorsal interossei and abductor pollicis brevis muscles. Abnormal spontaneous activity was not present in the left shoulder and arm (deltoid, biceps and triceps) and cervical paravertebral muscles. Intramuscular anterior scalene block did not reveal any improvement in symptoms. Patient was planned for conservative treatment followed by advanced radiological investigations in case of poor response to conservative therapy. Psychiatric consultation was taken to rule out psychosomatic dysfunction. Along with symptomatic medications, physical therapy was started with patient education, under the supervision of dedicated physiotherapist and occupational therapist. Patient compliance with home exercise program and modification of behavior pattern at home and at work was ensured by repeated counseling. Improvement in symptoms was noted by 2 months and >90% improvement described by the patient by 6 months. At the end of 1 year, patient was completely asymptomatic.

Discussion

There are a few reports of clavicular duplication in the literature.^[2-7] The first descriptions were incidental radiographic findings related to embryological phenomena.^[3-5] Golthamer reported the incidental finding of a supernumerary bone just



Figure 1: Maintained rounded contour of left shoulder with wasting. No obvious supraclavicular fullness



Figure 2: Left hand, a little pallor with wasting of hypothenar eminence. Abducted little finger can be noted due to weakness of 4th interossei

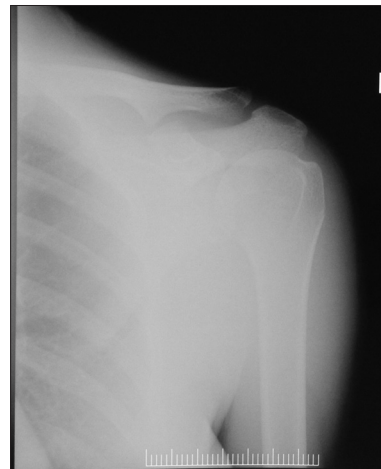


Figure 3: Radiograph showing duplication of left clavicle

below the clavicle and termed it “os subclaviculare.” He declared that clavicular duplication is a congenital anomaly and clinically insignificant.^[4] Twigg and Rosenbaum described duplication of the clavicle as “bifid” clavicle in a 40-year-old man. They opined that this was an anatomic variant, “without

clinical significance and purely of anatomic interest".^[3] Oestreich advocated that partial clavicular duplication was developmental, but that one variety of the lateral clavicle hook could be an acquired lesion or occur congenitally.^[5] Ogden suggested a traumatic etiology and was of the view that the early reports occurred due to the result of unrecognized injuries.^[6] The physis of the clavicle is inherently weaker than the acromioclavicular joint ligaments and trauma to the shoulder in skeletally immature (until late adolescence) results in fracture instead of acromioclavicular joint sprain and a "new" clavicle can form if the proximal clavicle remains displaced. The final result is a "duplicated" clavicle and new growth occurs from the distal epiphysis and toward the proximal diaphysis. Most importantly, all authors have agreed that duplicate clavicles are clinically insignificant.^[7]

The diagnosis of thoracic outlet syndrome can be made by history, physical examination, provocative tests, ultrasound, radiological evaluation, and electro-diagnostic evaluation. There are no reliable provocative tests, however certain provocative maneuvers, e.g., 90° abduction and external rotation (Roos test) has predictive value in screening. Roos described that light exercise in 90° abduction and external rotation causes maximum compression of the brachial plexus and subclavian vessels by narrowing the costoclavicular space.^[8,9]

This case is unique as association between clavicular duplication and thoracic outlet syndrome has been described: A fact hitherto unreported. Although, the bony causes of thoracic outlet syndrome described include abnormalities such as anomalous cervical ribs, hypoplastic first thoracic rib, exostoses of the first rib or clavicle and long transverse process of C7, no report of thoracic outlet syndrome due to duplication of clavicle was found on search of the literature.^[10,14] Radiographs should be advised to rule out this bony anomaly along with other common causes, as described above. Further clinical studies are required to establish the causation.

Conclusion

We conclude that, in cases of neurogenic thoracic outlet syndrome, radiograph of the shoulder region should be advised along with routine practice of the cervical spine radiography. One should be vigilant enough to rule out bony anomalies of

the clavicle in shoulder radiograph. Dedicated physiotherapy and occupational therapy should be offered to the patients of thoracic outlet syndrome for optimum management. Patients should be educated accordingly when this clavicular deformity is detected incidentally.

References

1. Dorskocil M. The development of the clavicle in man. *Gegenbaurs Morphol Jahrb* 1985;131:357-65.
2. Webb LX, Mooney JF. Fractures and dislocations about the shoulder. In: Green NE, Swiontkowski MF, editors. *Skeletal Trauma in Children*. 3rd ed. Philadelphia, PA: Elsevier Inc.; 2002. p. 322-5.
3. Twigg HL, Rosenbaum RC. Duplication of the clavicle. *Skeletal Radiol* 1981;6:281.
4. Golthamer CR. Duplication of the clavicle; (os subclavicular). *Radiology* 1957;68:576-8.
5. Oestreich AE. The lateral clavicle hook-an acquired as well as a congenital anomaly. *Pediatr Radiol* 1981;11:147-50.
6. Ogden JA. *Skeletal Injury in the Child*. 2nd ed. Philadelphia, PA: Elsevier Inc.; 1990. p. 441.
7. Wilson TC. Duplication of the clavicle. *Orthopedics* 2006;29:79-80.
8. Köknel Talu G. Thoracic outlet syndrome. *Agri* 2005;17:5-9.
9. Roos DB. Thoracic outlet syndrome is underdiagnosed. *Muscle Nerve* 1999;22:126-9; 137.
10. Rayan GM. Lower trunk brachial plexus compression neuropathy due to cervical rib in young athletes. *Am J Sports Med* 1988;16:77-9.
11. Roos DB. Congenital anomalies associated with thoracic outlet syndrome. Anatomy, symptoms, diagnosis, and treatment. *Am J Surg* 1976;132:771-8.
12. Nichols AW. Diagnosis and management of thoracic outlet syndrome. *Curr Sports Med Rep* 2009;8:240-9.
13. Atasoy E. Thoracic outlet syndrome: Anatomy. *Hand Clin* 2004;20:7-14.
14. Brantigan CO, Roos DB. Etiology of neurogenic thoracic outlet syndrome. *Hand Clin* 2004;20:17-22.

How to cite this article: Agarwal S, Akhtar MN. "Clavicular duplication causing thoracic outlet obstruction": Unique presentation of unreported association between clavicular duplication and thoracic outlet syndrome. *Ann Med Health Sci Res* 2014;4:317-9.

Source of Support: Nil. **Conflict of Interest:** None declared.