Bilateral coracoclavicular joints as a rare cause of bilateral thoracic outlet syndrome and shoulder pain treated successfully by conservative means

Singh V K, Singh P K, Balakrishnan S K

ABSTRACT

The coracoclavicular joint (CCJ) is a rare anomalous joint. Symptomatic CCJ, being an exceptional rarity, makes it difficult to formulate a standard set of practice or guidelines. We report a 50-year-old Indian man, a machine operator by profession, who experienced bilateral shoulder pain and arm paraesthesia for two years, and was diagnosed with bilateral CCJ. The symptoms gradually increased, affecting his daily activities. Dynamic magnetic resonance imaging revealed the compression of the brachial plexus in extreme shoulder abduction. After a thorough search of the literature, we retrieved four similar cases, all of them treated with individualised protocols. None of the cases was bilateral. The lack of clear evidence in any particular direction and the patient's medical condition prompted us to give a conservative trial, before embarking on more invasive methods. He showed rapid response to the conservative treatment with remission of all symptoms. To the best of our knowledge, this is the first reported case of bilateral symptomatic CCJ with bilateral thoracic outlet syndrome, that was managed conservatively.

Keywords: bilateral thoracic outlet syndrome, coracoclavicular joint, symptomatic coracoclavicular joint, thoracic outlet syndrome

Singapore Med J 2009;50(6):e214-e217

INTRODUCTION

The presence of a true synovial diarthrodial coracoclavicular joint (CCJ) is rare in humans.[1,2] Gruber was the first to describe the entity in 1861.[2] Since then, in spite of a reported incidence of 0.8%–1.2% of CCJ discovered in autopsy, radiological or osteological studies,[3,4] the number of symptomatic cases remain a rarity.[1] The incidental variety, although extensively studied, is of doubtful clinical significance. A painstaking literature search yielded four similar cases, none of which was bilateral. The current case is a unique example, where a bilateral CCJ led to the dynamic compression of the brachial plexuses by disturbing the normal shoulder mechanics.

CASE REPORT

A 50-year-old right-handed man of Indian origin, and a machine operator by profession, was referred to the orthopaedic clinic with a history of bilateral shoulder pain radiating to the arms. The pain was gradually increasing in severity over a period of two years, and was especially exacerbated by prolonged use of the arms. He had difficulty lifting heavy weights and had a loss of strength in the arm and grip. He had pins and needles radiating down the arm all the way to the fingers, particularly in the ulnar distribution (C8–T1). Similar symptoms were present on the left side as well, although of shorter duration. The pain became so severe on one occasion that he had to attend the emergency department for advice. There was no positive family history of associated shoulder, neck or rheumatological conditions. The initial impression from history was nerve root entrapment secondary to cervical spondylosis. On examination, the abduction was painful and limited to 100° in both shoulders. The brachialgia and paraesthesia were reproduced in extreme abduction on both sides. There was a slight loss of power (MRC grade 4/5) in grip in both hands. The examination of the cervical spine was unremarkable, with a full range of painless movement.

The patient was investigated further by imaging and nerve conduction studies. Radiographs of the shoulder showed well-formed accessory joints between the coracoid and distal clavicle bilaterally (Fig.1). The nerve conduction studies confirmed bilateral brachial plexus compression. The motor conduction velocity was decreased in the ulnar nerves to 56 m/sec (normal > 72 m/sec) across the thoracic outlet. Magnetic resonance (MR) imaging of both the shoulder joints, however, failed to reveal any compression of the brachial plexus

Department of Trauma & Orthopaedics Surgery, Luton and Dunstable Hospitals NHS Foundation Trust, Lewsey Road, Luton, LU4 0DZ, The United Kingdom

Singh VK, DOrth, MRCS, MCh
Registrar

Singh PK, MBBS, MS MRCS
Registrar

Balakrishnan SK, MBBS, MRCS
Trainee

Correspondence to:
Dr Vinay K Singh
Tel: (44) 79 6363 6753
Fax: (44) 15 8249 7403
Email: we.publish@googlemail.com

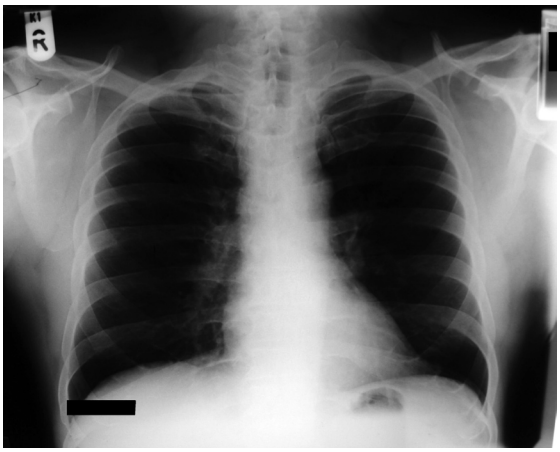

Fig. 1 Radiograph shows bilateral accessory coracoclavicular joints.

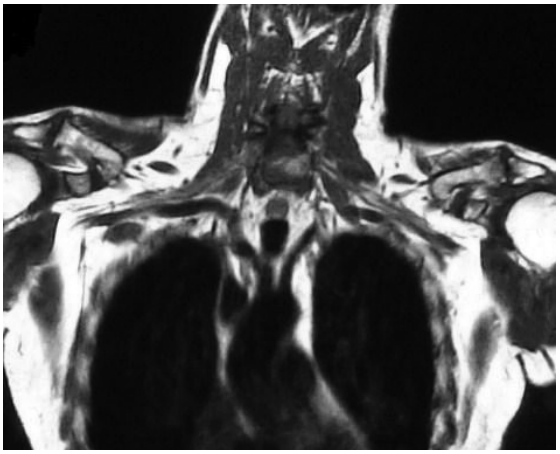

Fig. 2 Coronal T1-W MR image of both shoulders shows the well-formed coracoclavicular joints between the lateral end of clavicle and coracoid process bilaterally.

but substantiated the presence of bilateral well-formed CCJ (Fig. 2). The case was discussed in the fracture clinic with expert neuroradiologists. Due to the strong clinical and electrophysiological suspicion of brachial plexus compression, a dynamic MR imaging was performed. The dynamic MR imaging successfully delineated the underlying pathophysiology. The normal physiological elevation of clavicles during the abduction was reduced bilaterally. The restriction of the movement was most marked in the stage of extreme shoulder abduction. Compression of the brachial plexuses was noted bilaterally in the terminal stage of overhead abduction. Due to coexisting medical conditions and the patient's wish, he was managed conservatively. A good response to the conservative measures was seen, and he remained symptom-free at 12- and 18-month follow-up after discharge.

DISCUSSION

Most authors consider CCJ as a rare entity. Depending on the method of study and the population sample, the reported incidence in literature varies from 0.8% in large osteological studies to 1.2% in some radiological series.[3,4] However, most of these incidental discoveries rarely give rise to any symptom in an individual. After a painstaking literature search, we found four cases of CCJ with brachial plexus involvement (Table I). The first case was described by del Valle and Giordano in 1943.[5] They reported a white woman aged 35 years with a left shoulder pain. The pain gradually became worse in intensity with a radiation to left the mammary region, arm and neck. The second case was reported by Wertheimer in 1948.[6] The patient was a 37-year-old black manual worker, who presented with complaints of pain and limitation of movements in his left shoulder. Soon after the second case, a third case was reported by Hall in 1950.[7] The patient, a white male aged 40 years, was a talented organist. His symptoms started with pins and needles of the right hand and gradually progressed to frank pain. The pain was reproduced by a manoeuvre similar to Adson's test. More recently, in 1993, Hama et al described a fourth case of a 38-year-old female from Japan. This woman had a history of diffuse swelling and tenderness of the right coracoid process, along with pain and numbness in the right hand.[8] All these cases were diagnosed by radiographs of the shoulder region, which clearly outlined the presence of CCJ. Hama et al also performed arthrography of the anomalous joint.[8] Not surprisingly, owing to the non-availability of MR imaging at the time of the above-mentioned studies, it was not used by these authors.

In all these previous cases, the symptoms were ascribed to the compression of the brachial plexus. Three types of thoracic outlet syndromes have been described.[8] Cervical rib or scalenus syndrome, in which the compression of the neurovascular bundle occurs proximal to the clavicle; hyperabduction syndrome, in which the brachial plexus is being compressed distal to the clavicle; and costoclavicular syndrome, with a compression under the clavicle. del Valle and Giordano believed the symptoms to be sympathetic or plexal in origin due to the compression of microscopic nerves.[5] Hama et al suggested that apart from the anomalous CCJ, the abnormal attachment of the subclavius muscle also contributed to the compression of the brachial plexus in their case. The contraction of the subclavius muscle due to inflamed CCJ was thought to create a pressure on the neurovascular bundle. The claims were supported by the reversal of symptoms after the excision of anomalous joints.[8] In our case, the compression was more dynamic

Table I. Reported cases of CCJ with brachial plexus involvement.

Author	Year	No. of cases	Age(years) /gender	Clinical features	Side	Proposed pathology	Conservative trial	Management	Outcome
del Valle and Giordano[5]	1943	1	35/F	Left shoulder pain radiating to breast, arm and neck	Left	Compression of brachial plexus	Failed	Excision of CCJ by osteotomy via transdeltoid muscle approach	Complete resolution
Wertheimer[6]	1948	1	37/M	Shoulder joint pain radiating to left arm, limitation of shoulder movement	Left	Compression of brachial plexus	Failed	Excision of CCJ by osteotomy via transdeltoid muscle approach	Complete resolution
Hall[7]	1950	1	38/M	Pins and needles, numbness and pain - ulnar side of the forearm, weakness in the hand	Right	Compression of brachial plexus	Failed	Excision of CCJ by osteotomy via subclavian approach	Complete resolution
Hama et al[8]	1993	1	38/F	Swelling and tenderness right coracoid process, pins and needles in the right hand	Right	Compression of brachial plexus leading to thoracic outlet syndrome due to CCJ and subclavius muscle	Failed	Excision of CCJ by osteotomy via subclavian approach. Excision of subclavius muscle	Complete resolution
Singh et al (current case study)	2008	1	45/M	Shoulder pain, arm paraesthesias	Bilateral	Dynamic compression of bilateral brachial plexus	Successful	Rest, analgesics, lifestyle modification, physiotherapy	Complete resolution

in nature, seen only in extreme shoulder abduction.

The movements of a human shoulder represent the result of a complex dynamic interplay of many forces, allowing it a greater range of motion than any other joint in the body. An alteration in any of these factors may affect normal shoulder mechanics. Normally, the coracoclavicular ligament complex suspends the shoulder girdle from the clavicle at an average distance of 13 mm. The clavicle participates in all movements of the shoulder joint. During the shoulder abduction, the coracoclavicular ligament serves two important functions; it prevents the undue upward displacement of the clavicle,[9] but on the other hand, due to its inherent elasticity, it provides the clavicle sufficient freedom to permit reasonable physiological movement in an upward direction,[6] thus preventing the impingement of immediately underlying neurovascular structures. The presence of an anomalous coracoclavicular joint makes the clavicle "stiffer", reducing its normal range of movement.[10] The clavicle, therefore, can no longer enjoy the same freedom in the cephaled direction during shoulder abduction. It is proposed that the close proximity of the clavicle to the brachial plexuses may predispose them for compression in extreme abduction of the shoulder. The hypothesis was proven by the demonstration of brachial plexuses compression in dynamic MR imaging.

The asymptomatic cases are typically managed by masterly inactivity with a presumption of all of them being 'purely incidental'. However, in spite of having some evidence of predisposition to osteoarthritis by anomalous CCJ,[3] due to the extremely low incidence of symptomatic cases, a routine follow-up of incidental cases cannot be justified. The symptomatic group, however, requires active and clear management protocols. Diagnosis is usually apparent on radiographs. Sometimes when it is difficult to ascertain with surety the CCJ as a cause of the symptoms, the injection of local anaesthetic may be helpful in localising the cause.[6] All the cases described in the literature were managed surgically by the excision of the anomalous joint, after the failure of a conservative trial. Four authors used two different surgical approaches. While the authors of the first two studies, del Valle and Giordano, and Wertheimer, excised CCJ by an osteotomy performed via the transdeltoid muscle approach,[5,6] the authors of the latter two, Hall and Hama et al, did the same through the subclavicular approach.[7,8] A complete resolution of symptoms was achieved by all four studies. However, in our case, we decided to tailor the treatment in accordance to the patient's individual needs and circumstances. A complicated past medical history of deep vein thrombosis, pulmonary embolism and smoking, put him in ASA Grade 3, making surgery a less attractive option. The patient was reluctant to have any invasive procedure including local steroid injections. The decision of a conservative trial was logical especially in the presence of definitive precipitating factors, including physical

exertion and repeated overhead abduction. After an open discussion with the patient, we mutually decided to opt for conservative treatment. Due to the presence of clearly identifiable precipitating factors, the mainstay of treatment was rest and lifestyle modification. He was started on analgesics and anti-inflammatory medication, with an advice to take six weeks off from work, in order to ensure complete rest. A substantial improvement in the symptoms was noted in the first few weeks, with complete resolution in six weeks. The physiotherapy team was involved at this stage, and light mobilisation of both upper limbs was started. An occupational therapy (OT) input was sought for appropriate lifestyle modifications. The OT team taught him how to accomplish his day-to-day activities without much overhead abduction of the shoulder and had necessary adjustments made in his house as well. He ultimately changed to a less demanding job with no manual work involved.

To conclude, bilateral CCJ, though a rare entity, should be kept in the differential diagnosis of thoracic outlet syndrome once the common causes have been excluded. Whereas radiography remains a simple and inexpensive method of diagnosis, more sophisticated investigations, including electrophysiological studies and dynamic MR imaging, should be ordered in equivocal cases. In the presence of dynamic compression of the brachial plexus, simple measures like analgesics, rest and life-style modifications can be curative, avoiding the need for invasive interventions.

REFERENCES

1. Cheung TF, Boerboom AL, Wolf RF, Diercks RL. A symptomatic coracoclavicular joint. J Bone Joint Surg Br 2006; 88:1519-20.
2. Gruber WL. [A monograph with remarks including: contributions to the anatomy of the region infraclavicularis and deltoidea]. Mem Acad Sci St Petersbourg 1861; VII Series 3:1-28. German.
3. Gumina S, Salvatore M, De Santis R, Orsina L, Postacchini F. Coracoclavicular joint: osteologic study of 1020 human clavicles. J Anat 2002; 201:513-9.
4. Haas WH, Kingma MJ, Drucker F. The coracoclavicular joint and related pathological conditions. Ann Rheum Dis 1965; 24:257-66.
5. del Valle D, Giordano A. [Cervico-brachial pain syndrome caused by coracoclavicular articulation. Operation healing]. Rev Argent Norteam-Med Cine Med 1943; 1:687-93. Spanish.
6. Wertheimer LG. Coracoclavicular joint; surgical treatment of a painful syndrome caused by an anomalous joint. J Bone Joint Surg Am 1948; 30A:570-8.
7. Hall FJS. Coracoclavicular joint: a rare condition treated successfully by operation. Br Med J 1950; 1:766-8.
8. Hama H, Matsusue Y, Ito H, Yamamuro T. Thoracic outlet syndrome associated with an anomalous coracoclavicular joint. A case report. J Bone Joint Surg Am 1993; 75:1368-9.
9. Terry GC, Chopp TM. Functional anatomy of the shoulder. J Athl Train 2000; 35: 248-55.
10. Frassetto F. [Three cases of articulation coraro-clavicolare watchlist radiographically on Living]. Chir Organi Mov 1921; 5:116-124. Italian.