Axillary neuropathy mimicking quadrilateral space syndrome and its follow up for one year

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ABSTRACT

تعتبر الإصابة بالاعتلال العصبي للإبط نتيجة لتداخل في العصب في تجويف الربع الجانبي نادرة. وصفت حالة مريض يبلغ من العمر ٢٤عاماً تم تشخيصها باعتلال عصبي معزول في الإبط يشبه متلازمة تجويف الربع الجانبي. تم وصف ذلك في البداية بواسطة كاهيل وبالمر في عام ١٩٨٣م وتم تعريف ذلك بتداخل الفرع البعيد من عصب الإبط والشريان المقوس العضدي الخلفي في التجويف التشريحي ذو الأربع زوايا. تم متابعة المريض لعام كامل سريرياً ومن الناحية الفيزيائية الكهربائية. الهدف من التقرير من ذكر هذه الحالة هو التأكيد على أن الحالات التي لديها مثل هذه الطبيعة تمثل نوع من التحديات حيث تكون الحاجة إلى تدخل العديد من التخصصات الطبية للتشخيص والعلاج.

Axillary neuropathy due to entrapment of the nerve in the quadrilateral space is seen rarely. Here, we describe a 24-year-old patient diagnosed with isolated axillary neuropathy that mimicked quadrilateral space syndrome. Quadrilateral or quadrangular space syndrome (QSS), first described by Cahill and Palmer in 1983, was defined as the entrapment of the distal branch of the axillary nerve and the posterior humeral circumflex artery (PHCA) in the quadrangular shaped anatomic space. We tracked the follow up of the patient for one year both clinically and electrophysiologically. Our aim in reporting this case is to stress the point that cases of such a nature usually represent situations of diagnostic and treatment challenges, where multidisciplinary approaches are required.

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Quadrilateral or quadrangular space syndrome (QSS), first described by Cahill and Palmer in 1983[1] was defined as the entrapment of the distal branch of the axillary nerve and the posterior humeral circumflex artery (PHCA) in the quadrangular shaped anatomic space formed by the humerus and the teres minor, teres major, and long head of triceps muscles.[1] It is seen rarely and should be differentiated from painful weakness of shoulder abductors and external rotators, which comprises conditions that cause axillary nerve changes such as anterior shoulder dislocation, humeral neck fracture, brachial plexus stretch injuries, pressure from casts or splints, improper use of crutches, intramuscular injections, and brachial neuritis which is also called as Parsonage-Turner syndrome or neuralgic amyotrophy.[2] Rotator cuff tears and shoulder neuropathies may have similar clinical presentations. Pain, shoulder muscle atrophy, and limitation of motion are primary complaints in both. Distinguishing between the 2 is based on the history and physical examination.[2] Here, we report the one year follow-up of a patient with axillary neuropathy that mimicked QSS both clinically and electrophysiologically. In this report we aimed to stress that, although rarely seen, axillary neuropathy may represent a diagnostic and therapeutic challenge where a multidisciplinary approach is needed.

Case Report. A 24-year-old left handed farmer presented with left shoulder pain, weakness of shoulder girdle muscles, and numbness over the deltoid. The patient reported that his complaints began one month prior to his admittance, after carrying heavy sacks on his back and using various farming tools that required more strength than using the common ones. A story of a previous shoulder injury could not be detected. He reported pain that was poorly localized and always dull in character. Physical examination revealed weakness of shoulder abduction and external rotation besides atrophy of the left arm prominent on deltoid and teres minor muscles (**Figure 1**). The affected shoulder had normal range of motion with a stable glenohumeral joint. There was no scapular winging. The quadrilateral space was tender on palpation. The pain was aggravated

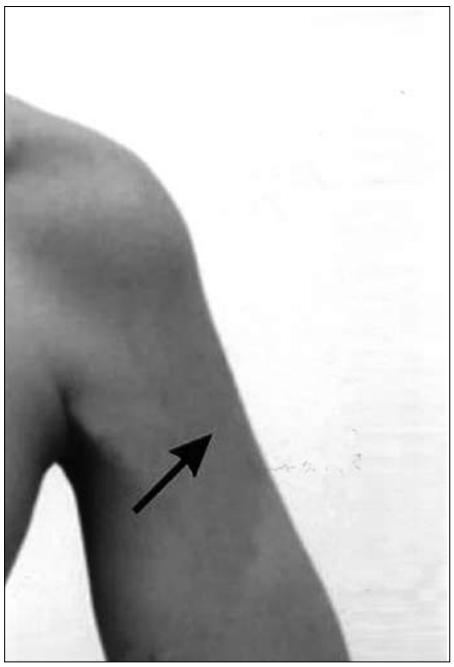

Figure 1 - Left deltoid atrophy.

with activity and improved with rest. Deep tendon reflexes were normal. There was no increase in deficit with compression of QS. An objective hypoesthesia and allodynia over the shoulder region were detected, but there was no sensory deficit on forearm or further muscle weakness, which could be indicative of a lesion of the brachial plexus. Axillary and radial pulses were normal and did not change after one minute of external rotation and abduction. Nerve conduction studies and needle EMG were performed (Neuropack 8-Nihon Kohden) after 2 days of the patient's admittance (32 days after his complaints began). Surface electrodes were used for both recording and stimulation, except when stimulating the suprascapular nerve where needle electrodes were used for recording from the supraspinatus muscle. In nerve conduction studies, left axillary nerve stimulation, recorded from the middle and posterior fibers of the deltoid muscle, according to the belly-tendon method, evoked compound muscle action potential (CMAP) responses with slightly long latencies and low amplitudes both with axillary and erb stimulations (**Table 1**). Recording from the anterior portion of deltoid evoked no CMAPs. Axillary nerve conduction studies were repeated to assure reproducibility. The motor nerve conductions of suprascapular, musculocutaneous, median, ulnar and radial motor nerves and the sensory nerve conduction studies including median, ulnar, radial superficial, and lateral antebrachial cutaneous nerves were in normal limits of our laboratory. On electromyography (EMG), a moderate degree of spontaneous activity comprising fibrillations and positive sharp waves was recorded both in the deltoid and teres minor muscles while recruitment was reduced in both of them. Durations of motor units were prolongated, and they were mostly polyphasic. Our patient fulfilled 3 out of 4 of the criteria suggested for the diagnosis of QSS, which were shoulder pain with undetermined localization, paresthesia over the shoulder, and lateral arm inconsistent with dermatomal distribution, and tenderness over the QS with palpation.[1] Although the clinical findings pointed out that the involvement of the axillary nerve could be just proximal to the QS or at the QS, MRI studies of the left shoulder and quadrilateral space were normal. Plain films of the shoulder also yielded normal results. The electrodiagnostic study could not locate the lesion to the QS, although it indicated very clearly that there was an axonal involvement of the left axillary nerve. The patient was diagnosed as an isolated axillary nerve injury by consultants from the departments of Orthopedic Surgery and Physical Medicine and Rehabilitation Unit. Physical therapy consisting of pendulum exercise, active range of motion, and a strengthening program was started for a painful active range of motion and shoulder girdle muscle weakness. The patient was to continue this exercise program at home for 6 months. He was invited for control every 2 weeks for the following 3 months and monthly for the next 6 months. Non-steroid antiinflammatory drugs and gabapentin were also initiated for his complaints. At the end of the first month after his admittance, his clinical and electrophysiological examinations did not change and the consultants advised surgical decompression of the axillary nerve in the QS, which was rejected by the patient. The clinical and electrodiagnostic follow-ups of second, third, and fourth months did not show any differences, either. At the sixth month, his physical examination changed slightly. The atrophy improved. He still had objective hypoesthesia on his shoulder, but no allodynia. Examinations of shoulder abduction and external rotation showed slight weakness. His electrodiagnostic examination did not reveal major differences from the 4 prior examinations, apart from the decrease in spontaneous activity of deltoid and teres minor muscles (**Table 2**). The following monthly examinations revealed slight improvements of strength of shoulder abductors and external rotators. His last physical examination was completely normal, the atrophy had significantly reduced, but he described a subjective feeling of numbness on his shoulder. He described no allodynia or any other symptom of neuropathic pain. Compression of the QS and abduction and external rotation of the shoulder no longer evoked pain. His seventh and last electrodiagnostic examination disclosed

Table 1 - Nerve conduction studies at admittance (32 days after the patient's first complaints began).

Nerve	Side	Recording	Latency	Normal	Amplitude (mv/μv)	Normal (mv/μv)	Distance (cm)	NCV	Normal
Axillary - axilla	Left	Deltoid	5.07	<4.5	1.75	>7.5			
Erb point			10.02	<8	1.83	>8.2	26	52.5	>58
Axillary - Erb point	Right	Deltoid	4.40	<8	26.1	>8.2	24	60	
Suprascapular - Erb point	Left	Supraspinatus	2.79	<3.1	29.4	>8			
Musculocutaneous - Erb point	Left	Biceps brachii	4.8	<4.9	15	>7.7			
Suprascapular - Erb point	Left	Infraspinatus	3.78	<3.9	19.5	>7.8			

NCV - Nerve conduction velocity, Normal - Our laboratory normal (of ages between 10 and 29)

Table 2 - Nerve conduction studies at the sixth month.

Nerve	Side	Recording	Latency	Normal	Amplitude (mv/μv)	Normal (mv/μv)	Distance (cm)	NCV	Normal
Axillary-axilla	Left	Deltoid	4.9	<4.5	2	>7.5			
Erb point			10	<8	1.9	>8.2	26	52	>58
Axillary - Erb point	Right	Deltoid	4.40	<8	26.1	>8.2	24	60	
Suprascapular - Erb point	Left	Supraspinatus	2.9	<3.1	27	>8			
Musculocutaneous - Erb point	Left	Biceps brachii	4.3	<4.9	16	>7.7			
Suprascapular - Erb point	Left	Infraspinatus	3.9	<3.9	20	>7.8			

NCV - Nerve conduction velocity, Normal - Our laboratory normal (of ages between 10 and 29)

Table 3 - Nerve conduction studies at the end of one year.

Nerve	Side	Recording	Latency	Normal	Amplitude (mv/μv)	Normal	Distance (cm)	NCV	Normal
Axillary - axilla	Left	Deltoid	4.00	<4.5	9	>7.5			
Erb point			7.50	<8	10	>8.2	26	74	>58
Suprascapular - Erb point	Left	Supraspinatus	2.5	<3.1	27 mV	>8			
Musculocutaneous - Erb point	Left	Biceps brachii	4.55	<4.9	18 mV	>7.7			
Suprascapular - Erb	Left	Infraspinatus	3.39	<3.9	19 mV	>7.8			

NCV -Nerve conduction velocity, Normal - Our laboratory normal (of ages between 10 and 29)

normal nerve conduction tests, although needle EMG of deltoid and teres minor muscles still showed signs of chronic denervation (**Table 3**).

Discussion. Although axillary neuropathy due to trauma is not a rare entity, QSS is rarely encountered. Most of the conditions mentioned above as differential diagnosis could be ruled out for our patient by history, physical examination, and normal imaging studies. Brachial plexus stretch injuries affect the supraclavicular plexus lesions and cause more extensive clinical neurological deficits. Parsonage-Turner Syndrome has a sudden onset, and pain is most commonly felt over the posterior-lateral deltoid. The pain awakens the patient at night, and the weakness is often patchy involving distal thumb flexion, pronation of forearm, and flexion of arm as well as the shoulder,[3] and it was quite impossible to explain our patient's situation with brachial plexus involvement, also. The QSS has been reported to be due to sporting activities such as volleyball, baseball, throwing athletism, prosthetic devices (with figure eight harness), lesions of paralabral cyst, glenoid labral cyst and ganglion, in one case with hereditary neuropathy with liability to pressure palsy and in one case with quadriplegia. The syndrome due to sporting activities is reported to be secondary to muscle hypertrophy, fibrous bands or contusion in the quadrilateral space.[4] There are only a small group of QSS cases whose electrophysiological data has been reported thoroughly, and the data differs quite significantly among them. Long term follow up for the electrophysiological findings of these patients is scarce in the literature. Our patient's nerve conduction studies showed an axonal involvement of the axillary nerve, and it is known that demyelination is seen in entrapment neuropathies. Although axonal involvement may become more prominent after early stages in entrapment neuropathies, prolongation of distal latencies always predominate and this was not the condition in our patient.

Some imaging studies of the QS by MRI, reported atrophy of the teres minor muscle as the most typical finding of QSS,[5] however, recent studies claim that this finding cannot be attributed to QSS alone.[6] It could not be detected in the QS MRI of our patient either. Arteriographic diagnosis of QSS is a point of debate and although it is suggested to be performed by some papers, there are papers also indicating that QSS results from compression of the axillary nerve, not from PHCA occlusion. Some authors also claim that arteriography is useless because of the high incidence of false positive results. In a study of MR angiography performed in healthy volunteers and in one patient with QSS, 80% of the healthy volunteers showed occlusion of the PHCA when their arms were hyper abducted and this finding was the same as the patient's. The authors concluded that MR angiography had no value in the diagnosis of the QSS.[7] We did not perform MR angiography in our patient.

Our patient described that his complaints occurred after a period of hard work. Just as is the case with sportsmen, we believe that there may be multiple causes. These may be stretching by excessive muscle activity during hard work or continuous compressions of the circumflex branch of the axillary nerve by the hypertrophic muscles. Muscular hypertrophy of the muscle boundaries of the QS may cause static and/or dynamic compression of the axillary nerve. The direct contusion effect of heavy sacks on the quadrilateral space or microtraumas to the nerve because of the long lasting muscle activity, especially on the dominant side may also play roles in establishing this condition. The space may be congenitally narrow, and it is known that nerves are more prone to entrapment while they pass through narrow spaces.

During the follow-up period, we noted that healing began after 6 months and was more prominent in the last 6 months. Whether this was the natural course of the neuropathic condition or a benefit from the rehabilitation program is unknown. In fact, it is not possible to exclude the natural history of the condition as being the result of the improvement, particularly with the lack of improvement at the early stages. The timing of the options for the treatment of QSS still remains controversial. Some authors favored surgical decompression as soon as the diagnosis was made, whereas, Baker and Liu[8] reported that initial treatment was usually nonoperative in neurovascular injuries to the shoulder. Surgery was usually reserved for patients suffering acute or chronic symptoms despite nonoperative treatment, and most patients were able to resume their previous activities in a timely manner with minimal disability. Helms[5] claimed that surgery for QSS was usually reserved for patients refractory to physical therapy. A recreational triathlete who had a spontaneous onset of QSS showed functional improvement within 6 weeks with a conservative rehabilitation program.[9] Steinmann et al,[10] reported that the vast majority of patients with axillary nerve injury recovered with non-operative treatment. On lack of clinical or electromyographic improvement, surgical options such as neurolysis, nerve grafting, or neurotization were recommended.

The diagnosis and treatment of isolated axillary neuropathies are multi-disciplinary, and electromyographers and neurologists dealing with peripheral nervous system should have knowledge of this syndrome, which may sometimes become a diagnostic challenge. Reports of clinical and electrodiagnostic

follow-ups of patients may help understand the nature of the involvement of axillary nerve, and which treatment options should be chosen.

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ILLUSTRATIONS, FIGURES, PHOTOGRAPHS

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