ACTA
ORTHOPAEDICA
et
TRAUMATOLOGICA
TURCICA

Acta Orthop Traumatol Turc 2009;43(2):190-192
doi:10.3944/AOTT.2009.190

Neuralgic amyotrophy as the primary cause of shoulder pain in a patient with rotator cuff tear

Rotator manşet yırtığı olan bir olguda omuz ağrısının ana nedeni: Nöraljik amiyotrofi

Ebru ŞAHİN, Özlem ŞENOCAK, A. Kadir BACAKOĞLU,[1] İbrahim ÖZTURA,[2] Mehtap GÖZÜM, Özlen PEKER

Departments of Physical Therapy and Rehabilitation, [1]Orthopedics and Traumatology, and [2]Neurology, Medicine Faculty of Dokuz Eylül University, İzmir

A 66-year-old woman with no history of trauma presented with severe shoulder pain. Magnetic resonance imaging revealed rupture of the supraspinatus tendon, for which surgical treatment was considered. However, it was noted that shoulder pain was accompanied by weakness in the shoulder muscles, and the patient underwent electroneuromyographic examination, which revealed neuralgic amyotrophy. Following physical therapy and rehabilitation combined with appropriate medical therapy, her symptoms significantly improved. In cases with severe shoulder pain without a trauma history, characteristics of pain should be thoroughly analyzed and neuralgic amyotrophy considered in the differential diagnosis.

Key words: Brachial plexus neuritis/therapy; diagnosis, differential; electromyography; rotator cuff/injuries; shoulder pain/etiology.

Öncesinde travma öyküsü olmayan 66 yaşında kadın hastada, şiddetli omuz ağrısı nedeniyle yapılan manyetik rezonans görüntülemede supraspinatus tendonunda tam kat yırtık saptandı ve hastaya cerrahi tedavi planlandı. Ancak, omuz ağrısına omuz kuşağında güçsüzlük eşlik etmesi nedeniyle yapılan elektronöromiyografik incelemede nöraljik amiyotrofi tanısı kondu. Uygulanan fizik tedavi ve rehabilitasyon programı ve medikal tedavi sonucunda hastanın semptomlarında belirgin gerileme oldu. Travmanın eşlik etmediği şiddetli omuz ağrılarında, ağrının özellikleri iyi değerlendirilmeli ve ayırıcı tanıda nöraljik amiyotrofi de düşünülmelidir.

Anahtar sözcükler: Brakiyal pleksus nöriti/tedavi; tanı, ayırıcı; elektromiyografi; rotator manşet/yaralanma; omuz ağrısı/etyoloji.

Detailed clinical features of neuralgic amyotrophy were described by Parsonage and Turner in 1948. Therefore, it has been known as Parsonage-Turner syndrome, acute brachial neuropathy, acute brachial plexitis, and idiopathic brachial plexopathy.[1] Acute and severe shoulder pain is the initial symptom, presenting neuropathic characteristics. Muscle weakness of the shoulder girdle follows pain. Clinical diagnosis is frequently confounded by shoulder and neck problems.[2] Underdiagnosis usually results in inappropriate treatment of the shoulder. We present a case of neuralgic amyotrophy accompanied by rotator cuff tear.

Case report

A 66-year-old female patient presented with persistent and burning left shoulder pain of three-month history, that increased in severity with motion. No trauma was reported. She underwent shoulder magnetic resonance imaging (MRI), which revealed a full-thickness rupture of the supraspinatus tendon. Her medical treatment was designed and surgery was planned. However, the patient presented again with a complaint of muscle weakness of the left shoulder. She was then referred to our clinic and hospitalized.

She had complaints of paresthesia, pain, and weakness. She had hypertension and was taking antihypertensive medication. The examination of the neck and right shoulder was normal. Physical examination of the left shoulder showed painful and limited range of motion. Active and passive range of motion testing showed the following: flexion 30° and 120°, abduction 30° and

Correspondence / *Yazışma adresi:* Dr. Ebru Şahin. Dokuz Eylül Üniversitesi Tıp Fakültesi, Fiziksel Tıp ve Rehabilitasyon Anabilim Dalı, 35340 Balçova, İzmir, Turkey. Tel: +90 232 - 412 39 68 e-mail: ebru.sahin@deu.edu.tr

Submitted / *Başvuru tarihi:* 15.07.2008 **Accepted / *Kabul tarihi:*** 23.02.2009

100°, internal rotation gluteal and lumbar 3, external rotation 25° and 35°, respectively. Muscle strength was measured as 2/5 in the left shoulder flexors, abductors, internal and external rotators, and elbow flexors, while other muscle strengths were normal. Atrophy was noted in the deltoid and supraspinatus muscles. Tendon reflexes were normal except for hypoactivity of the left biceps. There was no sensation deficit nor a pathological reflex. Cranial nerve examination was normal. Laboratory findings and plain radiography of the lung were normal. Her previous MRI scan showed full-thickness rupture of the supraspinatus tendon and acromiohumeral impingement. In cervical MRI, multisegmental disc pathologies were present, but there was no spinal cord compression. Electromyography (EMG) revealed denervation potential in the biceps brachii and deltoid muscles, and neurogenic motor unit potentials (MUP) in the supraspinatus and infraspinatus muscles. Cervical paraspinal muscles and other muscles innervated by the brachial plexus were normal. Median and radial sensory nerve conduction studies to the first digit showed decreased amplitude. Other sensory nerve conduction studies, motor nerve conduction in the median and ulnar nerves, and muscle action potential amplitude were normal. These findings indicated upper trunk involvement of the left brachial plexus and the diagnosis was made as neuralgic amyotrophy.

The patient received physical therapy and rehabilitation for the left shoulder, including transcutaneous electrical nerve stimulation (TENS), hot pack, ultrasound, and exercise program (range of motion exercises, stretch exercises, and exercises with a continuous passive motion machine). The patient's pain was unresponsive to 1500 mg/day paracetamol and 150 mg/day diclofenac sodium. Considering neuropathic characteristics of the shoulder pain, gabapentin was added to her medical treatment and the dosage was gradually increased to 1800 mg/day. She received 20 sessions of physical therapy and rehabilitation program. The patient showed remarkable improvement after medical and physical therapy. The severity of pain decreased and passive range of motion returned to normal range. Active range of motion increased by 30° in all directions. There was no change in muscle strength. She was discharged from hospital on gabapentin (1800 mg/day) and with recommendation of home exercises. Follow-up examinations showed significant pain relief, so the dosage of gabapentin was gradually decreased. At sixth-month visit, she complained of no pain and gabapentin was discontinued. Active and passive range of motion values were as follows: flexion 130° and 180°, abduction 110° and 180°, internal rotation lumbar 3 and thoracal 12, and external rotation 45° and 50°, respectively. Muscle strength was measured as 3+/5 for shoulder flexion and abduction, 4/5 for external rotation, 4+/5 for internal rotation, and 3/5 for elbow flexion. Control EMG showed increases in radial and median sensory potential amplitudes and regeneration MUPs in the deltoid, supraspinatus, and infraspinatus muscles.

She was evaluated again in the twelfth month of follow-up. She had no pain. Range of motion was measured as flexion 150°/180°, abduction 140°/180°, internal rotation lumbar 2/thoracal 11, and external rotation 50°/60°. Muscle strength was 5/5 in shoulder flexors, abductors, external, and internal rotators, and 4/5 in elbow flexors. Electromyography showed chronic neurogenic MUP alterations and increase in the number of MUPs.

Discussion

Shoulder pain is a common complaint in patients presenting to the clinics of orthopedics and physical therapy and rehabilitation. In the differential diagnosis of shoulder pain, causes other than shoulder problems must be considered. Especially neuralgic amyotrophy must be considered in the differential diagnosis of patients with acute shoulder pain without a history of trauma. The incidence of neuralgic amyotrophy was reported as 1.64/100.000.[3] Although trauma is the most common cause of brachial plexopathy, in a case series of 203 patients, Moghekar et al.[4] found that neuralgic amyotrophy was the most common cause (%40). Although it can be seen at all ages, it occurs most frequently in the 3rd and 7th decades. Males are more commonly affected than females.[1]

Idiopathic and hereditary forms of neuralgic amyotrophy have been described. The idiopathic form can develop after infections, immunization, intensive exercise, and surgery; however, a specific etiologic factor may not be found.[1] In our case, there were no previous symptoms nor a family history or a history of infection, immunization, intensive exercise, or surgery. Her laboratory findings were also normal. No possible cause of neuralgic amyotrophy could be derived.

Diagnosis of neuralgic amyotrophy is based on medical history, physical examination, radiographic findings, and electrophysiologic studies.[3] Electromyography performed at least 3 to 4 weeks after the onset of symptoms is helpful in locating the lesion and differential

diagnosis.[3,5] Electrophysiologic studies are also helpful in determining the extent of denervation and subclinical involvement of the contralateral side.[6] The brachial plexus can be affected totally or partially. Isolated involvement of the upper extremity nerves can also be seen. Most affected nerves are the long thoracic and anterior interosseous nerves.[7] Cranial nerve involvement can be seen.[3] On the MRI scan, abnormal neurogenic high T2 signal intensity and atrophy can be seen in the affected shoulder girdle muscles. These findings are not specific to neuralgic amyotrophy, but support the diagnosis.[8] Laboratory examinations are usually normal.[3]

Treatment of neuralgic amyotrophy is symptomatic. The treatment aims to relieve pain, prevent restriction of movement by range of motion exercises, and restore muscle strength.[3,9] There is no evidence that one treatment modality is superior to another.[9] Corticosteroid and immunoglobulin therapies have been recommended in the early period of disease to prevent progression of muscle weakness, but effectiveness of these therapies has not been proven.[1,3] Analgesics and nonsteroidal anti-inflammatory drugs are recommended for pain relief. In our case, initial treatment with these drugs was not effective, so gabapentin (1800 mg/day) was started, which resulted in regression of pain in the early period. To our knowledge, use of gabapentin has not been reported in the treatment of neuralgic amyotrophy, but we thought that it might be helpful in this case because of the neuropathic characteristic of pain. Literature data on prognosis are variable. Although a favorable prognosis is usually mentioned,[6,10] van Alfen et al.[5] reported persisting pain and paresis up to three years in two-thirds of patients with neuralgic amyotrophy. Upper trunk involvement of the plexus has a better prognosis than that of lower trunk involvement.[3] Recurrences can be seen in 1-5% of patients (especially in hereditary form and phrenic nerve involvement).[3,11] In our case, the upper trunk was involved and the symptoms substantially regressed within a year.

The presented patient had symptoms which began with shoulder pain without muscle weakness. The characteristics of her pain was acute, severe, and burning without a history of trauma. Shoulder MRI revealed full-thickness rupture of the supraspinatus tendon, and her symptoms were initially attributed to this condition. As the weakness of the shoulder girdle and biceps muscles developed, the patient was assessed with EMG and the findings were consistent with neuralgic amyotrophy. Rotator cuff problems must be considered in the differential diagnosis of neuralgic amyotrophy. However, this patient had coexisting neuralgic amyotrophy and rotator cuff problems and MRI findings complicated the diagnosis. Therefore, characteristics of pain must be considered in the diagnosis of patients with shoulder pain. Neuropathic characteristic and distribution of pain must be warning and require further examination. Neuralgic amyotrophy must be kept in mind in the diagnosis of patients with burning pain and other neurologic problems must be eliminated. Since muscle weakness can also accompany chronic rotator cuff problems, a detailed neurological examination will be helpful in the differentiation of a brachial plexus lesion and cervical problems. As neuralgic amyotrophy falls within the scope of several disciplines, timely diagnosis may prevent unnecessary investigations and invasive approaches. Moreover, informing the patient about the good prognosis of the disease will decrease the patient's anxiety and increase adherence to treatment.[3]

References

1. Sathasivam S, Lecky B, Manohar R, Selvan A. Neuralgic amyotrophy. J Bone Joint Surg [Br] 2008;90:550-3.
2. Hosey RG, Rodenberg RE. Brachial neuritis: an uncommon cause of shoulder pain. Orthopedics 2004;27:833-6.
3. Rubin DI. Neuralgic amyotrophy: clinical features and diagnostic evaluation. Neurologist 2001;7:350-6.
4. Moghekar AR, Moghekar AR, Karli N, Chaudhry V. Brachial plexopathies: etiology, frequency, and electrodiagnostic localization. J Clin Neuromuscul Dis 2007;9:243-7.
5. Van Alfen N, van Engelen BG. The clinical spectrum of neuralgic amyotrophy in 246 cases. Brain 2006;129(Pt2):438-50.
6. Cruz-Martínez A, Barrio M, Arpa J. Neuralgic amyotrophy: variable expression in 40 patients. J Peripher Nerv Syst 2002;7:198-204.
7. Preston DC, Shapiro BE, editors. Brachial plexopathy. In: Electromyography and neuromuscular disorders: clinical-electrophysiologic correlations. 2nd ed. Philadelphia: Butterworth-Heinemann; 2005. p. 479-500.
8. Gaskin CM, Helms CA. Parsonage-Turner syndrome: MR imaging findings and clinical information of 27 patients. Radiology 2006;240:501-7.
9. Mamula CJ, Erhard RE, Piva SR. Cervical radiculopathy or Parsonage-Turner syndrome: differential diagnosis of a patient with neck and upper extremity symptoms. J Orthop Sports Phys Ther 2005;35:659-64.
10. Tsairis P, Dyck PJ, Mulder DW. Natural history of brachial plexus neuropathy. Report on 99 patients. Arch Neurol 1972; 27:109-17.
11. Lahrmann H, Grisold W, Authier FJ, Zifko UA. Neuralgic amyotrophy with phrenic nerve involvement. Muscle Nerve 1999;22:437-42.