Case Report / Olgu Sunumu

Idiopathic Brachial Neuritis Mimics Cervical Radiculopathy: A Case Report

İdiyopatik Brakiyal Nörit Servikal Radikülopatiyi Taklit Eder: Olgu Sunumu

Evrim Karadağ Saygı[1], Canan Şanal Toprak[2], Koza Çubukçu[1], Hakan Gündüz[1]

[1]Marmara University Faculty of Medicine, Department of Physical Medicine and Rehabilitation, İstanbul, Turkey
[2]Horasan State Hospital, Clinic of Physical Medicine and Rehabilitation, Erzurum, Turkey

Abstract

Idiopathic brachial neuritis (IBN) is an uncommon clinical disorder with an onset of acute, aching shoulder pain, followed by progressive muscle weakness and atrophy of the shoulder girdle and upper extremity. A 47-year-old male patient presented with an onset of acute left shoulder pain, which has lasted for two months. On physical examination, he also demonstrated a severe upper extremity weakness. Imaging tests showed a compression at C6 level on the left foraminal canal. According to these findings, the primary diagnosis was cervical radiculopathy, which may have required surgical intervention. However, the detailed history of pain, physical examination findings and electroneuromyography (ENMG) results were consistent with IBN that has a slow but progressive recovery with conservative treatment. Although a detailed history and physical examination are key points in the diagnosis of IBN, ENMG is the most helpful method for early and correct diagnosis that helps avoid unnecessary interventions.

Keywords
Brachial neuritis, cervical radiculopathy, electrophysiology

Öz

İdiyopatik brakiyal nörit (İBN) akut başlangıçlı omuz ağrısını, omuz kuşağı ve üst ekstremitede gelişen progresif kas güçsüzlüğü ve atrofinin takip ettiği nadir görülen bir hastalıktır. Kırk yedi yaşında erkek hasta sol kol ve omuzda ani başlayan ve 2 aydır devam eden şiddetli ağrı şikayeti ile başvurdu. Ayrıca üst ekstremitede fizik muayene ile gösterilebilen ciddi kas güçsüzlüğü vardı. Manyetik rezonans görüntülemede C6 seviyesinde foraminal kanalda basısı mevcuttu. Bu bulgulara göre ön tanı cerrahi girişim gerektirebilecek servikal radikülopati idi. Ancak detaylı anamnez, fizik muayene bulguları ve elektronöromiyografi (ENMG) sonuçları konservatif tedavi ile yavaş ancak progresif iyileşme gösteren İBN ile uyumlu idi. İBN tanısında kilit noktalar detaylı anamnez ve fizik muayane olsa da gereksiz girişimleri engellemek amacıyla erken ve doğru tanı için ENMG en faydalı testtir.

Anahtar Kelimeler
Brakial nörit, servikal radikülopati, elektrofizyoloji

Received/Geliş Tarihi : 29.04.2016
Accepted/Kabul Tarihi : 09.08.2016

doi:10.4274/meandros.galenos.2016.2822

Address for Correspondence/Yazışma Adresi:
Evrim Karadağ Saygı Assistant Professor, Marmara University Faculty of Medicine, Department of Physical Medicine and Rehabilitation, İstanbul, Turkey
Phone : +90 532 361 23 07
E-mail : evrimkaradag4@hotmail.com

ORCID ID: orcid.org/0000-0002-7420-9902

Introduction

Idiopathic brachial neuritis (IBN) is a peripheral neuropathy, with an unknown etiology, which mostly affects the upper trunk of brachial plexus, although focal involvement of individual nerves is also seen. Although there is no consensus about the most frequently affected nerve, suprascapular, long thoracic, radial, phrenic and axillary nerves were reported as commonly involved nerves in literature (1,2). This clinical entity is also known as brachial neuralgia, brachial plexus neuropathy, neuralgic amyotrophy, acute brachial radiculitis and Parsonage-Turner syndrome. It has a reported incidence of 1/1000/year (3). It is 30-50 times higher than previously thought because of the expanding clinical awareness of IBN (3). This syndrome usually affects adults between ages 20-60. Typical clinical course starts with unilateral shoulder pain followed by an augmented response to painful stimuli. Motor weakness develops approximately within days to 2 weeks and may be seen in an irregular distribution due to patchy involvement of brachial neuritis (1,4). Although detailed physical examination and considering IBN in differential diagnosis are the key points, electroneuromyography (ENMG) is the most helpful study in diagnosis.

Case Report

Informed consent statement was obtained from the patient for this study. A 47-years-old male patient presented with an acute onset left shoulder and arm pain which has been lasting for two months. On physical examination, manual muscle testing of supraspinatus, infraspinatus, deltoid, biceps and triceps on the left side were graded 1/5. Sensory deficit was not detected. Left brachioradialis and biceps deep tendon reflexes were diminished. There was no restriction in the passive range of motion of left shoulder however pain was exacerbated with the shoulder movement. Shoulder pain was present but decreased as compared his initial severe pain. Past medical history was unremarkable. His laboratory findings like erythrocyte sedimentation rate, complete blood count and routine biochemical examinations were within normal limits. C-reactive protein and rheumatoid factor were negative.

In cervical magnetic resonance imaging (MRI) on level C5-6 there was left posterolateral-foraminal disc herniation and probable compression on left C6 root

Table 1. Nerve conduction studies

	Latency (ms)	Amplitude (µV)	Velocity (m/sn)
Median (L, motor)	2.75	6.5	-
	6.50	6.3	59.7
Ulnar (L, motor)	1.95	6.72	-
	5.35	7.44	58.8
	7.1	6	51.4
Median (L, sensory)	2.58	42.9	50.4
Ulnar (L, sensory)	1.88	51	58.5
Lateral antebrachial cutaneous (L, sensory)	SAP was not obtained		
Median (R, motor)	3	9.2	-
	6.2	8	58.5
Ulnar (R, motor)	2.05	6	-
	5	6.8	62.3
	6.05	7	58.3
Median (R, sensory)	2.44	50.7	57.4
Ulnar (R, sensory)	2.02	46.3	54.5
Lateral antebrachial cutaneous (R, sensory)	3.28	12.7	52.2

L: Left, R: Right, ms: Milliseconds, SAP: Sensory action potential

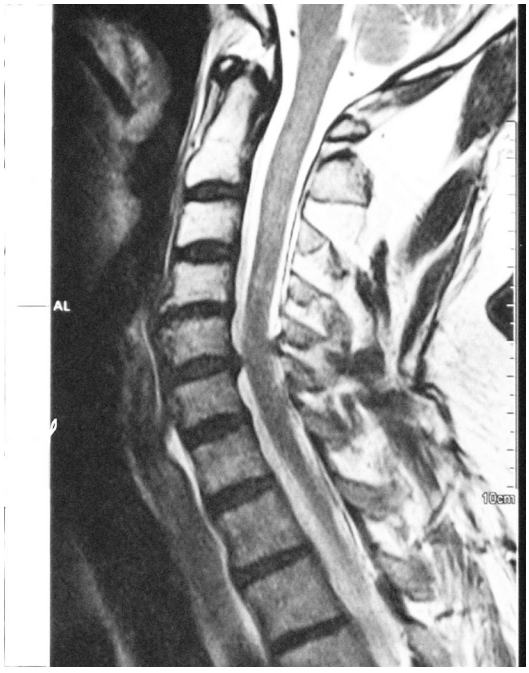

Figure 1. Sagittal cervical magnetic resonance imaging on level C5-6

(Figure 1, 2). In the electrophysiological study bilateral median, ulnar and radial nerve conduction studies were normal, but on the left side lateral antebrachial sensory nerve action potentials were absent (Table 1). In needle electromyography (EMG), there were abnormal findings in spontaneous action potentials in deltoid, biceps, triceps, brachioradialis and 1st dorsal interosseous muscles bilaterally. Although both extremities were affected, pathological findings were more prominent on the left side. On left deltoid muscle motor unit action potentials were absent. Cervical paraspinal muscle studies were within normal limits (Table 2).

The patient was diagnosed as IBN by evaluating the type of pain, diffuse muscle weakness and ENMG results. Gabapentin and non-steroidal anti-inflammatory drugs were prescribed to reduce inflammation of brachial nerves and pain, as recommended in literature (2,5). Physical therapy programme was designed. After two months his pain was relieved and muscle weakness was improved slightly. When ENMG was repeated after three

Table 2. Needle electromyography findings

Side	Muscle	Spontaneous Activities		MUP analyzing			Interference
		PSW	Fibrillation	Duration	Ampl	Polyphase	
L	Deltoid	++	++	Voluntary MUP was not obtained			
L	Supraspinatus	Ø	Ø	N	↑	↑	Mildly decreased participation
L	Biceps	+++	+++	↑↑↑	↑	↑	Severely decreased participation
L	Triceps	++	+	↑	↑	↑↑↑	Severely decreased participation
L	Brachioradialis	++	++	↑	↑	↑↑	Single oscillation
L	1.Dorsal interosseous	+	+	N	N	↑	Mildly decreased participation
L	APB	Ø	Ø	N	N	N	Submaximal
R	Deltoid	++	++	↑	N	↑	Mildly decreased participation
R	Triceps	+	+	N↑	N	N↑	Mildly decreased participation
R	1.Dorsal interosseous	++	++	N↑	↑		Mildly decreased participation
L	Paraspinal C6	Ø	Ø				

L: Left, R: Right, PSW: Positive sharp wave, MUP: Motor unit potential, Ampl: Amplitude, APB: Abductor pollicis brevis, N: Normal, ↑: Increased , Ø: No spontaneous activity potentials, +: Spontaneous activity potentials persistent in at least two areas, ++: Moderate number of persistent spontaneous activity potentials in 3 or more áreas, +++: Large number of persistent spontaneous activity potentials in all áreas, +++: Profuse, widespread, persistent spontaneous activity potentials that fill the baseline

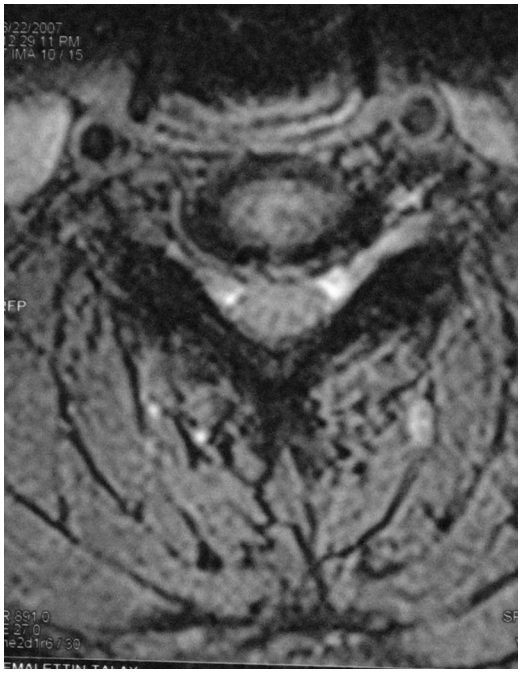

Figure 2. Axial cervical magnetic resonance imaging on level C5-6

months, partial regeneration findings were evident. On clinical examination pain and muscle weakness were detected also on the asymptomatic side.

Discussion

This patient was admitted to physical medicine and rehabilitation clinic with an acute severe left shoulder pain, which decreased in the last weeks. He also complained about progressive upper extremity weakness, demonstrated on physical examination.

In this case, primary diagnosis was cervical radiculopathy (CR). Compression in C6 level in MRI, on the left foraminal canal was evident. However, the patients' symptoms were also consistent with typical clinical course of IBN. Therefore, a differential diagnosis between CR and IBN was planned to avoid unnecessary interventions, because of their similar features, particularly at the time of onset.

CR and IBN are characterized by pain in shoulder, and upper extremity. Although CR generally has a gradual onset, IBN has a sudden onset of severe pain. Progression of upper extremity weakness is generally related to increasing pain in CR patients, while upper extremity weakness related to IBN typically develops as the pain decreases. While neck movements generally aggravate the symptoms of CR, the symptoms of IBN worsen with shoulder movement (6). In this case, presence of the pain exacerbated by shoulder movement and also upper extremity weakness related to reducing pain were consistent with IBN.

The prominent symptom is the pain in shoulder girdle. Pain is severe, burning and sharp; it radiates up to neck, down to forearm on the lateral side. Pain is acute and night pain is prominent (7,8). It may last from a couple of hours to 2-3 weeks.

Muscle weakness and pain symptoms may be seen together, but shoulder pain followed by patchy muscle weakness is a more common profile (4,9). In the series of van Alfen et al. (8), which consists of 246 cases, weakness developed within 24 hours in 33%, 1-7 days in 40%, 1-2 weeks in 14% cases. Weakness does not always follow the same route that pain follows. Atrophy may develop in the affected muscles (10).

Mild and patchiness sensory symptoms occur in a majority of patients on careful examination, frequently in the distribution of the lateral cutaneous antebrachial nerve. Sensory or motor symptoms may not match the distribution of pain and may not be noticed by patient because of the severe pain; it is very important clue for diagnosis of IBN (5,11). In this case, there was no clinically confirmed sensory loss, even though antebrachial sensory action potentials were absent in the ENMG study, and this was considered as an evidence of IBN.

In the cases of IBN, laboratory findings are within normal range. Complete blood count, sedimentation rate, serum electrolytes, liver enzymes, the chemical analysis of urine samples and immunological studies are within normal limits. (4,9). In this case laboratory findings proved no abnormalities, as mentioned. However, laboratory findings are also expected within normal range in CR patients and don't have a role to differentiate IBN from CR.

When considering the differential diagnosis, musculoskeletal disorders of shoulder, CR, poliomyelitis, amyotrophic lateral sclerosis, herpes zoster, tumors of spinal cord or brachial plexus, posterior interosseous nerve palsy, traumatic or compressive nerve injuries should be considered (12,13).

In this case ENMG study was planned to make the differential diagnosis between CR and IBN.

ENMG can localize the lesion and help confirm the diagnosis (4,9,14). Nerve conduction studies of the motor nerves are generally within normal limits. The amplitude of sensory nerve potentials may be smaller than expected. Needle EMG is the most helpful study in this diagnosis. In the needle EMG of proximal muscles with weakness and atrophy, fibrillation and positive sharp waves are evident (6). Paraspinal studies are usually in normal limits. It usually seems unilateral but about a third of these patients have bilateral involvement with an asymmetrical severity. Hence, even though the patient may complain of unilaterally clinical symptoms, bilaterally ENMG findings may be seen (15).

According to literature it is said that electrophysiological findings of C6 radiculopathies are less typical and specific compared to other radiculopathies on different levels. C6 radiculopathies may have electrophysiological symptoms of C5 and/or C7 radiculopathies (9). This case had diffused proximal muscle weakness and showed denervation findings on EMG studies. These symptoms may have been due to C6 radiculopathy. However, it was unlikely to explain the fibrillation and positive sharp waves of first dorsal interosseous muscle, which is innervated by C8-T1 roots. Patchy distribution findings of paresis supported diagnosis of IBN. On the other hand, this case had also unilaterally clinical symptoms but bilaterally ENMG findings that were consistent with bilateral IBN. Lateral antebrachial cutaneous sensory responses were absent, and this is a symptom of upper trunk brachial plexopathy. Paraspinal EMG results were normal as would be expected with IBN. When all of the results above were considered these led us to the diagnosis of IBN.

IBN is a benign disorder with good prognosis. Approximately 80%-90% of the patients recover after 2-3 years, but 35%-70% may remain with residual paresis and exercise intolerance (16,17). Complete recovery of muscle strength and functioning is common. However, duration of recovery alters from one patient to the next. It may go from 1 month up to 3 years. Sensory and motor improvements occur at the same rate. If pain is intense and muscle weakness is severe, healing process takes more time. If there is more than one nerve involved, recovery is expected to be slower. In upper trunks lesions, prognosis is better (10,16). Exceptionally spontaneous recovery does not take place and the patient may require surgical intervention.

To conclude, incidence of IBN is not as rare as previously thought. It needs to be considered as a differential diagnosis in patients who complain about acute intense shoulder pain. A detailed history and physical examination are required for diagnosis. CR should be excluded to avoid unnecessary and inappropriate interventions. In order to differentiate IBN from CR, detailed ENMG studies on both affected and unaffected (contra-lateral) sides may help clinicians for early diagnosis.

Ethics

Informed Consent: Informed consent statement was obtained from the patient for this study.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: E.K.S., H.G., Design: E.K.S., K.Ç., Data Collection or Processing: C.Ş.T., K.Ç., E.K.S., Analysis or Interpretation: H.G., C.Ş.T., E.K.S., K.Ç., Literature Search: H.G., C.Ş.T., E.K.S., K.Ç., Writing: H.G., C.Ş.T., E.K.S., K.Ç.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Crag A, Richardson JK. Rehabilitation of patients with neuropathies. In: Braddom RL editor. Physical Medicine and Rehabilitation. 4th ed. : Elsevier Saunders; 2011: 1065-95.
2. Monteiro Dos Santos RB, Dos Santos SM, Carneiro Leal FJ, Lins OG, Magalhães C, Mertens Fittipaldi RB. Parsonage-Turner syndrome. Rev Bras Ortop 2015; 50: 336-41.
3. van Alfen N, van Eijk JJ, Ennik T, Flynn SO, Nobacht IEG, Groothuis JT, et al. Incidence of neuralgic amyotrophy (Parsonage Turner syndrome) in a primary care setting--a prospective cohort study. PLoS One 2015; 10: e0128361.
4. Miller JD, Pruitt S, McDonald TJ. Acute brachial plexus neuritis: an uncommon cause of shoulder pain. Am Fam Physician 2000; 62: 2067-72.
5. Van Eijk JJ, Groothuis JT, Van Alfen N. Neuralgic amyotrophy: An update on diagnosis, pathophysiology, and treatment. Muscle Nerve 2016; 53: 337-50.
6. 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.
7. Gonzalez-Alegre P, Recober A, Kelkar P. Idiopathic brachial neuritis. Iowa Orthop J 2002; 22: 81.

8. van Alfen N, van Engelen BG. The clinical spectrum of neuralgic amyotrophy in 246 cases. Brain 2006; 129: 438-50.
9. Dumitru D, Zwarts M. Brachial plexopathies and proximal mononeuropathies. In: Dumitru D, Amato A, Zwarts M. Electrodiagnostic medicine. 2nd ed. St Luis: Mosby; 2002: 623-4.
10. Moghekar AR, Moghekar AR, Karli N, Chaudhry V. Brachial plexopathies: etiology, frequency, and electrodiagnostic localization. Journal of clinical neuromuscular disease 2007; 9: 243-7.
11. van Alfen N. The neuralgic amyotrophy consultation. J Neurol 2007; 254: 695-704.
12. Fransz DP, Schönhuth CP, Postma TJ, van Royen BJ. Parsonage-Turner syndrome following post-exposure prophylaxis. BMC Musculoskeletal Disord 2014; 15: 265.
13. Yang JS, Cho YJ, Kang SH, Choi EH. Neuralgic Amyotrophy Manifesting as Mimicking Posterior Interosseous Nerve Palsy. J Korean Neurosurg Soc 2015; 58: 491-3.
14. Simmons Z. Electrodiagnosis of brachial plexopathies and proximal upper extremity neuropathies. Physical medicine and rehabilitation clinics of North America 2013; 24: 13-32.
15. Saurez GA. Immune brachial plexus neuropathy. In: Dyck P, Thomas PK. editor. Peripheral Neuropathy. 4th ed. : Saunders; 2005: 2299-308.
16. Tjoumakaris FP, Anakwenze OA, Kancherla V, Pulos N. Neuralgic amyotrophy (Parsonage-Turner syndrome). J Am Acad Orthop Surg 2012; 20: 443-9.
17. van Alfen N. Clinical and pathophysiological concepts of neuralgic amyotrophy. Nat Rev Neurol 2011; 7: 315-22.