

Diabetic Cervical Radiculopathy with Adhesive Capsulitis of the Shoulder

Jeong-Hwan Seo and Sung-Hee Park

Department of Physical Medicine and Rehabilitation, Chonbuk National University Medical School, Chonbuk, Korea.

The common form of diabetic neuropathy is symmetrical peripheral polyneuropathy, which involves the distal part of the lower extremities whereas diabetic amyotrophy is seen in the proximal part of the lower extremities. Although other regions may also be affected, the presence of upper extremity involvement has rarely been emphasized. Diabetic radiculopathy may involve the cervical region before, after, or concurrently with lumbosacral radiculopathy. We report 2 rare cases of diabetic radiculopathy which involves the cervical region without involving the lumbosacral region. To our knowledge, these are the first reported cases of diabetic radiculopathy involving the cervical region only. In our cases, severe adhesive capsulitis in a shoulder was noticed together with cervical radiculopathy. Both diabetic radiculopathy and adhesive capsulitis have a poorly understood pathogenesis and their combined presence is presumed to be rare. Clinical features and management of cervical radiculopathy with adhesive shoulder capsulitis in 2 diabetic patients is described.

Key Words: Diabetic radiculopathy, cervical radiculopathy, adhesive shoulder capsulitis

INTRODUCTION

Diabetes mellitus (DM) is the most common metabolic disease.^{1,2} Several distinct types of peripheral neuropathy are associated with DM, such as distal symmetric sensory or sensorimotor polyneuropathy, autonomic neuropathy, diabetic neuropathic cachexia, polyradiculoneuropathies,

cranial neuropathies and other mononeuropathies.

Diabetic radiculopathy, also known as diabetic amyotrophy, is the most common syndrome of an asymmetric, subacutely progressive weakness and pain affecting the lower extremities.³ In 1953, Garland and Taverner, under the heading of "diabetic myelopathy" described a primary motor syndrome in the legs of 5 diabetic patients.⁴ Garland coined the noncommittal descriptive term "diabetic amyotrophy" because of uncertainty regarding involvement of the spinal cord in this syndrome.⁵ Other regions, however, may also be affected. Several reports have documented thoracolumbar radiculopathies that occur alone or accompany lower extremity pain and weakness.^{6,7} In contrast, the presence of upper extremity involvement, as a feature of this disorder, has barely been reported.⁸ In this report, two clinical experiences of diabetic cervical radiculopathy combined with adhesive shoulder capsulitis are described.

CASE REPORT

Patient 1

A 45-year-old man was admitted to hospital with a history of weakness of the left upper extremity and restricted range of motion in the left shoulder for 6 months. He was diagnosed as type 2 DM 3 months previously and medicated with an oral hypoglycemic agent. There was no history of antecedent illness, trauma, or other potential immunological triggers in this patient. The strength of left shoulder flexion, extension,

Received May 28, 2003

Accepted September 2, 2003

Reprint address: requests to Dr. Sung-Hee Park, Department of Physical Medicine and Rehabilitation Medicine, Chonbuk National University Medical School, 634-18, Keumam-dong, Dukjin-gu, Jeonju, Chonbuk 561-712, Korea. Tel: 82-63-250-1795, Fax: 82-63-254-4145, E-mail: shpark0130@hanmail.net

and abduction was 3/5. The range of motion of the left shoulder was limited in various aspects flexion 100°, extension 60°, abduction 30°, adduction 25°, with hard feelings at the end range which were suggestive of the profound adhesion of the shoulder joint. Reflexes were normal. Sensation was intact in bilateral upper extremities. Inspection revealed a winging scapular of the left shoulder and atrophy of the biceps, supraspinatus, and infraspinatus muscles of the left side (Fig. 1). Fasting blood glucose level ranged from 140 to 250 mg%. The glycosylated hemoglobin level was elevated to 7.7%. Serum muscle enzyme, serum creatinine and chest roentgenograms were all normal and serum viral markers were negative. The patient was referred for magnetic resonance imaging (MRI) of the cervical spine to rule out a herniated nucleus pulposus as the origin of his symptoms. MRI showed a herniated nucleus pulposus at the C5-6 level of the right paramedian side focally, but no evidence of disc herniation on the left side (Fig. 2). Arthrography of the left glenohumeral joint

showed an absent axillary pouch and diminished injection volume of contrast material in the joint cavity. These arthrographic findings were compatible with adhesive shoulder capsulitis (Fig. 3). Electrophysiologic examination of the upper limb, lower limb, cervical and lumbar paraspinal muscles was performed. Nerve conduction studies showed normal finding in the bilateral upper and lower extremities.

Needle electromyography demonstrated abnormal spontaneous activity in the left paracervical, supraspinatus, infraspinatus, deltoid and biceps muscles and motor unit firing was rapid and recruitment was reduced. These findings were indicative of C5 radiculopathy of the left side. The patient was treated with comprehensive rehabilitative treatment including physical therapy. Manual manipulation of the shoulder for the treatment of the frozen shoulder was performed under general anesthesia and an intraarticular injection with local anesthetics and steroid was administered into the glenohumeral joint. The range of motion of the shoulder was restored

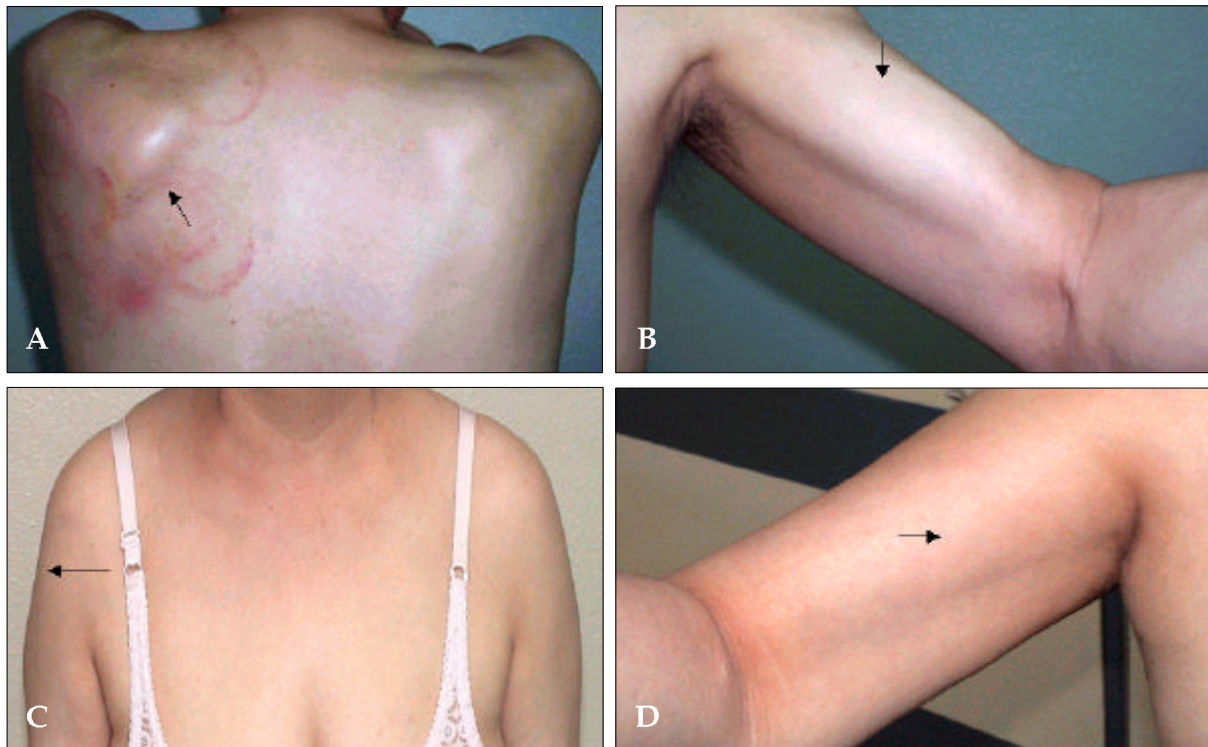


Fig. 1. Photographs of patients. Winging scapula and atrophic change of the left supraspinatus, infraspinatus (A) and left biceps brachii (B) were seen in patient 1. The Photograph of patient 2 shows atrophic change of the right deltoid (C) and biceps brachii muscles (D).

immediately after the manipulation. The follow-up examination at 1 month after discharge revealed the strength of the shoulder to be above 4/5. There was no evidence of proximal neuropathy, weakness or pain of the limbs during 6 months of follow-up examination.

Patient 2

A 48-year-old woman was admitted to hospital with weakness and sensory loss of the right upper extremity and a limited range of motion in the right shoulder which was refractory to conventional treatment over a 10 month period. She had been diagnosed type 2DM and has been medicated with oral hypoglycemic agent since 1997. The patient had no other history of trauma and illness. The strength of the flexion, extension, abduction and adduction of the right shoulder was 3/5. The range of motion of the right shoulder was limited in various aspects, flexion 90°,

extension 30°, abduction 60°, adduction 0°, internal rotation 0° and external rotation 40°. There was impaired sensation to pinpricks over the lateral side of the right arm. There was muscular wasting of the right deltoid and biceps (Fig. 1). Fasting blood glucose level ranged from 140 to 280 mg%. The glycosylated hemoglobin level was elevated to 8.4%. Serum muscle enzyme, serum creatinine and chest roentgenograms were all normal and serum viral markers were negative. The MRI of the cervical spine showed no evidence of abnormal findings. Arthrography of the right glenohumeral joint showed poor filling of the axillary pouch and increased thickening of the joint capsule up to 6 mm (Fig. 3). Magnetic resonance imaging of the right shoulder showed a small axillary pouch and an irregular articular margin (Fig. 4). Nerve conduction studies showed normal values in the bilateral upper and lower extremities. Needle electromyography demonstrated abnormal spontaneous activities such as fibrillation and positive sharp waves in the right paracervical, supraspinatous, infraspinatous, deltoid and biceps muscles as well as large amplitude, polyphasic motor unit potential in these muscles. These findings were indicative of the C5 radiculopathy of the right side. The patient was refractory to comprehensive rehabilitative treatment including physical therapy with thermal agent and manual therapy. Therefore treatment with intraarticular corticosteroid injection in conjunction with manipulation under general anesthesia was instituted. The patient was admitted to hospital for 3 weeks for this procedure. On discharge, the range of shoulder movement was improved in flexion, extension, abduction, adduction, and internal and external rotation. At the follow-up examination at 6 months post dis-

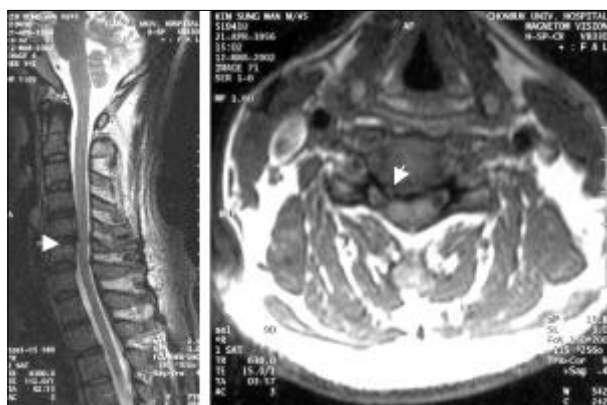


Fig. 2. Magnetic resonance image of patient 1 shows a herniated nucleus pulposus at the C5-6 level on the right paramedian side focally (white arrow) but no evidence of disc herniation on the left side.



Fig. 3. Arthrogram of the left shoulder in patient 1 shows a small axillary pouch (A). The arthrography of the right glenohumeral joint showed poor filling of the axillary pouch and increased thickening of the joint capsule up to 6 mm in patient 2 (B).

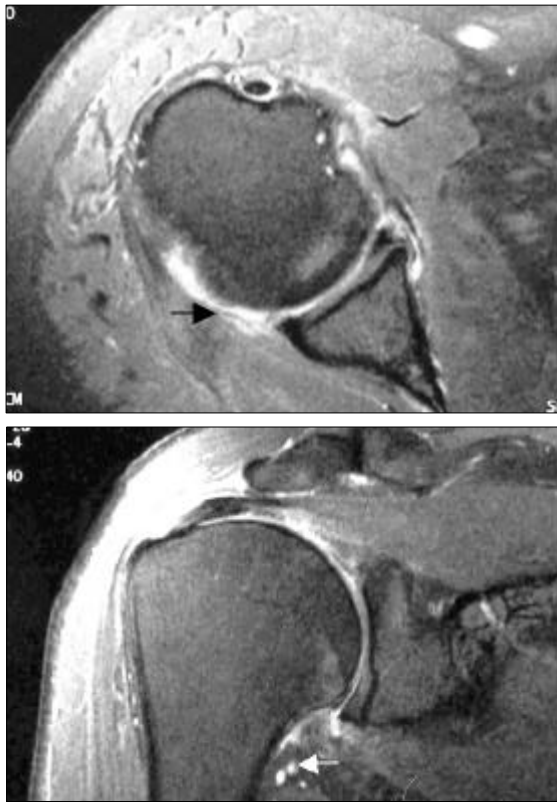


Fig. 4. Magnetic resonance image of the right shoulder in patient 2 shows an irregular articular margin (black arrow) and small axillary pouch (white arrow).

charge, the strength and the range of motion in that shoulder had improved to near normal range. Follow up electromyography revealed that there was no electrophysiologic evidence of cervical radiculopathy, thoracolumbar radiculopathy or mononeuropathy.

DISCUSSION

Patients with type 2 DM may experience any of the various types of diabetic neuropathies.² Several previous studies have mentioned arm involvement associated with painful, asymmetric, proximal diabetic neuropathies using various designations, but to date there has been no clear consensus regarding the nature of the upper extremity syndrome.³ Proximal diabetic neuropathy may be a heterogeneous syndrome, and we should recognize a broad variability in clinical manifestations of this illness. Raff and Asbury

were the first to emphasize that diabetic amyotrophy could be associated with more widespread involvement of individual peripheral nerves.⁹ More recently Dyck et al and Katz et al reported upper limb radiculoplexopathy with diabetic lumbosacral radiculoplexopathy.^{1,3} However, these patients had proximal upper limb involvement that began simultaneously with the lower limb disorder. This study describes two patients with diabetic radiculopathy, who electrophysiologically demonstrated involvement of the C5 nerve root. In these cases, there was no known neuropathic symptom or sign in a lower extremity. To the knowledge of these investigators, there has been no report of diabetic radiculopathy only involving a cervical root.

Diabetic neuropathy is probably not a single pathogenetic entity. It is comprised of different syndromes, each with a distinct clinical presentation and possible multifactorial pathogenetic mechanisms such as metabolic defects, ischemic lesions or a combination of these two factors. The pathogenesis of proximal diabetic neuropathy remains uncertain. The absence of obvious changes in the nerve conduction study and quantitative autonomic tests, before and after the onset of proximal diabetic neuropathy may indicate that the pathogenesis of this condition is different from distal symmetrical diabetic polyneuropathy.^{10,11} Recently, several investigators have reported that this syndrome is attributable to epineuria or perineurial microscopic vasculitis, due to neuropathologic findings in biopsied nerve fibers.^{10,11} The C5 nerve root was affected in the 2 cases of this study.

Considering the involvement of the C5 root among other cervical roots in these cases, the C5 root appears to be particularly prone to an ischemic insult or other pathogenic process of DM. Although the mechanism is unclear, C5 nerve root involvement is also common in giant cell arteritis which is systemic vasculitis.¹² Preferential involvement of C5 nerve roots may depend on the vascularization of cervical roots.

The etiology of adhesive capsulitis remains unclear. Trauma, autoimmune disorders, cervical dysfunction, radiculopathy, tendinitis, bursitis, and hormonal changes have all been proposed as possible precipitating factors. According to the

present illness history of the cases in this study, adhesive capsulitis has seemed to develop as a complication of the neuropathic process, a cervical radiculopathy involving C5 nerve root.

The treatment of the patient with adhesive capsulitis involved the use of thermal agents, manual therapy, continuous passive motion and splinting of the arm in an elevated position. Manipulation of a frozen shoulder was reported to offer reduced pain and a shorter period of disability in patients who fail to respond to conservative treatment of frozen shoulder syndrome. In the cases in this study, a prompt increase in shoulder mobility and decrease in shoulder symptoms followed manipulation under anesthesia with steroid injection into the shoulder joint.

The majority of cases of diabetic radiculopathy recover fully within 3 years.¹³ In others, recovery may be incomplete depending on the degree of wasting and the distance of the lesion from the site of injury. In the 2 case studies reported here, the major concern of treatment was management of the adhesive capsulitis and the outcome was satisfactory when manipulation was done under anesthesia.

According to the experience of these investigators, when cervical radiculopathy is complicated in DM patients, it can proceed to serious adhesive shoulder capsulitis. Thus it is suggested that much attention and preventive measures should be given to these patients.

REFERENCES

1. Dyck PJ, Kratz KM, Karnes JL, Litchy WJ, Klein R, Pach JM, et al. The prevalence by staged severity of various types of diabetic neuropathy, retinopathy, and nephropathy in a population-based cohort: The Rochester diabetic neuropathy study. *Neurology* 1993;43:817-24.
2. Ross MA. Neuropathies associated with diabetes. *Med Clin North Am* 1993;77:111-24.
3. Katz JS, Saperstein DS, Wolfe G, Nations SP, Alkhesam H, Amato AA, et al. Cervicobrachial involvement in diabetic radiculoplexopathy. *Muscle Nerve* 2001;24:794-8.
4. Garland H, Taverner D. Diabetic myelopathy. *Br Med J* 1953;1:1405-8.
5. Garland H. Diabetic amyotrophy. *Br Med J* 1955;2:1287-90.
6. Kikia DG, Breuer AC, Wilbourn AJ. Thoracic root pain in diabetes: the spectrum of clinical and electromyographic findings. *Ann Neurol* 1982;11:80-5.
7. Sun SF, Streib EW. Diabetic thoracoabdominal neuropathy: clinical and electrodiagnostic features. *Ann Neurol* 1981;9:75-9.
8. Barohn RJ, Sahenk Z, Warmolts JR. The Bruns-Garland syndrome (diabetic amyotrophy). Revisited 100 years later. *Arch Neurol* 1991;48:1130-5.
9. Raff MC, Asbury AK. Ischemic mononeuropathy and mononeuropathy multiplex I diabetes mellitus. *N Engl J Med* 1968;279:17-22.
10. Said G, Goulon-Goeau C, Lacroix C, Moulouguet A. Nerve biopsy findings in different patterns of proximal diabetic neuropathy. *Ann Neurol* 1994;35:559-69.
11. Sail G, Lacroix C, Lozeron P, Ropert A, Plante V, Adams D. Inflammatory vasculopathy in multifocal diabetic neuropathy. *Brain* 2003;126:376-85.
12. Rivest D, Brunet D, Desbiens R, Bouchard JP. C-5 radiculopathy as a manifestation of giant cell arteritis. *Neurology* 1995;45:1222-4.
13. Tsairis P, Dyck PJ, Mulder DW. Natural history of brachial plexus neuropathy report on 99 patients. *Arch Neurol* 1972;27:109-17.