CASE REPORT

Lumbosacral Plexus Injury and Brachial Plexus Injury Following Prolonged Compression

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We report the case of a 36-year-old woman who developed right upper and lower limb paralysis with sensory deficit after sedative drug overdose with prolonged immobilization. Due to the initial motor and sensory deficit pattern, brachial plexus injury or C8/T1 radiculopathy was suspected. Subsequent nerve conduction study/electromyography proved the lesion level to be brachial plexus. Painful swelling of the right buttock was suggestive of gluteal compartment syndrome. Elevation of serum creatine phosphokinase and urinary occult blood indicated rhabdomyolysis. The patient received medical treatment and rehabilitation; 2 years after the injury, her right upper and lower limb function had recovered nearly completely. As it is easy to develop complications such as muscle atrophy and joint contracture during the paralytic period of brachial plexopathy and lumbosacral plexopathy, early intervention with rehabilitation is necessary to ensure that the future limb function of the patient can be recovered. Our patient had suspected gluteal compartment syndrome that developed after prolonged compression, with the complication of concomitant lumbosacral plexus injury and brachial plexus injury, which is rarely reported in the literature. A satisfactory outcome was achieved with nonsurgical management. [*J Chin Med Assoc* 2006;69(11):543–548]

Key Words: brachial plexus injury, compartment syndrome, lumbosacral plexus injury

Introduction

Brachial plexus injuries are usually associated with multisystem trauma. These injuries can range from palsies that recover spontaneously to avulsion of the nerve roots with no return of function.[1] Upward traction with internal rotation and abduction of the arm mostly results in lesions of lower cervical nerve roots. It is generally recognized that limited avulsion of the lower roots usually has a better prognosis.[1] While the majority of brachial plexus injuries are associated with traction injuries, there are reports of brachial plexus injury resulting from direct compression or hematoma.[1,2] Lumbosacral plexopathies can be divided into those caused by structural lesions and those caused by nonstructural lesions. Clinical presentation includes motor and sensory deficits in distribution of the multiple nerves originating from the plexus.[3] Acute lumbosacral plexus injury is rare and is usually associated with retroperitoneal hematoma, hemophilia, stabbing injury, surgical operation, dislocation of the hemipelvis, or pelvic fracture.[4] Among the various causes of lumbosacral plexus injury, gluteal compartment syndrome can be potentially debilitating. Elevation of gluteal compartment pressure in the nondistensible osseofascial boundaries can cause ischemia and cellular damage in associated muscles and nerves, which may result in massive rhabdomyolysis, renal failure, multiple-organ failure and death.[5] Due to its clinical rarity in the literature, we herein present a case of concomitant brachial and lumbosacral plexus injures after prolonged compression with suspected compartment syndrome. Permanent disabilities were prevented through early intervention with rehabilitation.

Case Report

A 36-year-old female patient was referred to the emergency department with persistent right upper

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and lower limb weakness and sensory loss. She had been found lying on the floor approximately 12 hours after she was seen taking hypnotics and alcohol. On awakening, she experienced weakness of her right leg and hand, associated with increasing pain and swelling in her right thigh and buttock.

On arrival in the emergency room, physical examination showed right-side limb weakness, more severe in the 4th and 5th finger flexors, abductors of the thumb, index and little finger, and the whole lower limb. Motor testing using the Medical Research Council (MRC) 6-point scale revealed weakness in right finger abduction (MRC grade 2), right finger flexion (MRC grade 3), right knee flexion (MRC grade 3/5), right hip extension (grade 3/5), right ankle dorsiflexion (grade 0/5), and right ankle plantar flexion (grade 0/5). Sensory deficit was found over the medial side of the right upper arm, forearm, 4th and 5th fingers, and the whole right lower limb, especially the below-knee area. The patient's right buttock was massively swollen, mottled, and tender. Deep tendon reflexes in her right-side limbs were absent. Popliteal and pedal pulses were symmetric bilaterally. However, no compartment pressures were obtained.

Laboratory investigation revealed white blood cell count of 14,000/mm^3 with neutrophils predominantly high, creatinine phosphokinase of 1,173 U/L, creatine kinase isoenzyme MB of 26 U/L, creatinine of 0.9 mg/dL, blood urea nitrogen of 27 mg/dL, and urinary occult blood of 3+ with 2–4 red blood cells/high power field. Musculoskeletal sonography showed right lower limb subcutaneous edema with patent bilateral femoral veins and popliteal veins. Computed tomography (CT) of the pelvis showed marked soft tissue swelling of the right gluteal muscles with diffuse subcutaneous lymphedema and infiltration over bilateral abdominal walls, pelvis, and bilateral upper thighs (Figure 1).

The patient was admitted and treated with antibiotics for right thigh cellulitis. Alkaline hydration was administered for presumptive rhabdomyolysis. Follow-up musculoskeletal sonography 2 weeks later revealed right gluteal muscle swelling with heterogeneous echogenicity. Electromyography/nerve conduction study (EMG/NCS) performed 43 days after the injury showed no response over the right median, ulnar, peroneal, tibial and sural nerves. Needle EMG of the right upper limb revealed active denervation over the dorsal interossi, abductor pollicis brevis, and sternal part of the pectoralis major, and decreased recruitment over the above muscles were still noted (Table 1). Therefore, brachial plexus injury at the medial cord or lower trunk was diagnosed. Needle EMG of the right lower limb showed active denervation over the anterior tibialis, gastrocnemius, rectus femoris, and adductor magnus. No active denervation sign was found over the tensor fascia lata and gluteus maximus (Table 2). Therefore, right lumbosacral plexus lesion was diagnosed.

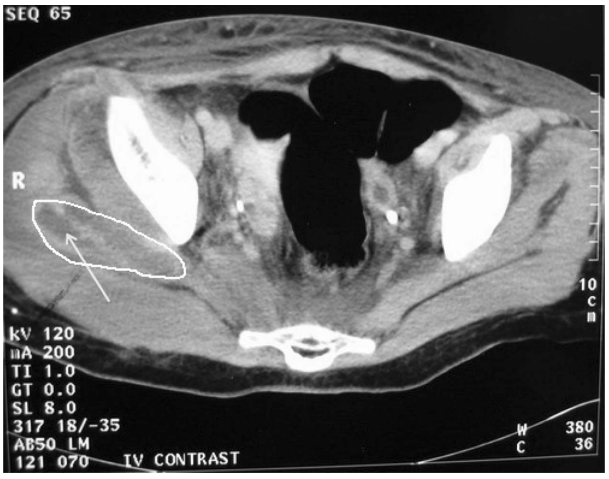

Figure 1. Prominent soft tissue swelling with suspected intramuscular cyst formation over right-side gluteal muscles (arrow).

The rehabilitation program included gentle range-of-motion exercises for the affected limb joints, electrical stimulation applied over the anterior tibialis, gastrocnemius, and quadriceps, strengthening exercises that emphasized muscle stabilization, balance training, and ambulation training. The patient was discharged with assistance of a quadricane when walking.

Two years after discharge, the patient could walk independently with nearly full recovery of her right hand function. However, she continued to suffer from right side foot drop. Knee extensors and flexors had recovered partially (MRC grade 4/5). EMG/NCS showed decreased amplitude over the left peroneal, tibial and sural nerves, with active denervation signs over the anterior tibialis and gastrocnemius. Increased polyphasic motor unit action potentials over muscles, including vastus medialis, adductor longus, gluteus maximus and gluteus medius, were observed, indicating ongoing nerve regeneration in these specific muscles. However, the patient refused further EMG/NCS study of her right upper limb.

Discussion

Acute lumbosacral plexus injury is rare and usually associated with retroperitoneal hematoma and trauma.[3] The lumbosacral plexus is thought to be less prone to injury because it is afforded greater protection by the

Table 1. Electromyography of the upper limbs 43 days after injury

Muscle (right)	Spontaneous activity		MUAP	Recruitment
	Fibrillation	Positive sharp wave		
Dorsal interossi	++	++	Normal	Poor
Abductor pollicis brevis	++	++	Normal	Poor
Brachioradialis	–	–	Normal	Full
Extensor digitorum communis	–	–	Normal	Full
Biceps	–	–	Normal	Full
Triceps	–	–	Normal	Full
Deltoid	–	–	Normal	Full
Sternal part of pectoralis major	+	+	Normal	Poor

MUAP = motor unit action potential.

Table 2. Electromyography of the lower limbs 43 days after injury

Muscle (right)	Spontaneous activity		MUAP	Recruitment
	Fibrillation	Positive sharp wave		
Anterior tibialis	++	++	No volitional potential	
Gastrocnemius	+	+	No volitional potential	
Rectus femoris	+	+	No volitional potential	
Extensor digitorum brevis	+	+	No volitional potential	
Adductor magnus	+	+	No volitional potential	
Tensor fasciae latae	–	–	Normal	Rich
Gluteus maximus	–	–	Normal	Rich

MUAP = motor unit action potential.

pelvic bones.[4] The rate of nerve injury following pelvic fracture is reported to range from 0.75% to 12.0%.[4] Characteristic clinical findings of lumbosacral plexopathy may include pain and motor and sensory deficits in several areas of the lower limbs depending on the segment of lumbosacral plexus involved. The important point is that the area of motor or sensory deficit should not be distributed in a specific nerve or root supplied pattern. Diagnosis and evaluation of the lumbosacral plexus may be made by electrodiagnostic testing, magnetic resonance imaging, or CT.[4] In electrodiagnosis, denervation changes should be found in muscles of at least 2 different nerve roots and of at least 2 different peripheral nerves with sparing of the paraspinal muscles.[4] In our patient, lumbosacral plexus injury was diagnosed solely by electrodiagnostic study, in which active denervation signs were noted over the anterior tibialis and gastrocnemius, but not over the tensor fascia lata or gluteus maximus. In addition, sural sensory nerve action potential, which is supposed to be intact in radiculopathy, could not be obtained. Therefore, root lesion could be excluded. The patient's pelvic CT scan showed no evidence of pelvic fracture or hematoma. Therefore, the actual etiology was possibly due to prolonged compression resulting in muscle damage and edema.

Brachial plexus injury is common in traumatic injury. The usual symptoms of brachial plexus injury include paralysis of the shoulder, arm, and/or hand with paresthesia and altered sensation.[1] The mechanism of injury to the brachial plexus is either from extreme traction on the nerves or direct impact. Downward traction generally results in lesions on the upper cervical nerve roots, whereas upward traction results in lesions of lower cervical nerve roots such as C8 and T1.[1] In the case we have presented, EMG study showed active denervation over the dorsal interossi, abductor pollicis brevis, and sternal part of the pectoralis major, which are supplied by the ulnar nerve, median nerve, and medial pectoral nerve, respectively. Due to the low possibility of concurrent injuries to these 3 nerves, the differential diagnoses could only be medial cord injury, lower trunk injury or C8/T1 radiculopathy. However, C8/T1 root lesions could be excluded based on the unobtainable sensory response. Therefore, injury at the medial cord or lower trunk was diagnosed. Although we could not obtain data on the patient's posture during prolonged lying, we may assume that

the patient lay with her right arm in the abducted position, which is a position prone to cause lower trunk or medial cord lesion. The characteristic findings of lower trunk or medial cord lesion include sensory deficit over the medial side of the upper arm, forearm, and hand, which are supplied by the medial brachial cutaneous nerve, medial antebrachial cutaneous nerve, and ulnar nerve, respectively. Motor weakness in the muscles supplied by the medial pectoral nerve, ulnar nerve and part of the median nerve, including the abductor pollicis brevis, flexor pollicis longus, and 1st and 2nd lumbricals, should also be present.

The distinctive feature of this case lies in the concurrent involvement of the sensory and motor function of the right-side limbs, which can also be observed in patients with Brown-Sequard syndrome spinal cord injury or stroke. The key to differentiating between plexus injury and spinal cord injury or stroke lies in successfully distinguishing upper from lower motor neuron lesions. In upper motor neuron lesions such as spinal cord injury or stroke, deep tendon reflexes should be increased. Pathologic reflexes such as Babinski's sign or Hoffmann's sign should be positive. Increased spasticity should also be present over the involved muscles instead of flaccid status. Furthermore, in the case of Brown-Sequard syndrome spinal cord injury, the area of sensory deficit supplied by the spinothalamic tract should be on the opposite side of the area of motor deficit. In addition, specific motor weakness pattern with relative sparing of other muscles, as often occurs in plexus injury patients, should not be present in stroke or spinal cord-injured patients.

Concurrent lesions of the brachial plexus and lumbosacral plexus are extremely rare in the literature. Kanter et al[6] reported a patient with diffuse large B-cell lymphoma who presented with unilateral progressive peripheral sensorimotor neuropathy, which was later diagnosed as lumbosacral and brachial neurolymphomatosis. The patient was successfully treated by intensive chemotherapy and autologous stem cell transplantation. Neuralgic amyotrophy is a well recognized disorder mimicking plexopathy due to involvement of multiple nerve-supplied muscles but sparing paraspinal muscle.[7] However, neuralgic amyotrophy usually attacks with severe neuralgic pain with minimal sensory loss, and motor deficit often develops after the pain subsides. Neuralgic amyotrophy is usually limited to the brachial plexus, and concurrent lumbosacral involvement is rare. Inoue et al[8] reported a 23-year-old woman with idiopathic brachial and lumbosacral plexopathy who presented with a relapsing course of weakness and numbness following acute onset of pain in the right shoulder or pelvic girdle area; she was successfully treated with low-dose intravenous immunoglobulin. Marra[7] reported a 42-year-old man with clinical and neurophysiologic evidence of recurrent neuralgic amyotrophy involving both the brachial and lumbosacral plexus. In spite of spontaneous recovery in 6 weeks, the patient initially had poor response to medical treatment, including prednisolone, carbamazepine and codeine. Acute systemic involvement of the nervous system such as acute inflammatory demyelinating polyneuropathy, porphyria, diphtheria, drugs (dapsone, nitrofurantoin, vincristine), toxins (arsenic, thallium, triorthocresyl phosphate), tick paralysis or vasculitis should also be considered in concurrent weakness over upper and lower limbs, but they are usually bilateral, which was not the case in our patient.

Another clinical symptom of note in our patient was right painful, swollen buttock. Elevated creatine phosphokinase and urinary occult blood with 2–4 red blood cells/high power field indicated rhabdomyolysis.[9] Although there was no actual demonstration of gluteal compartment pressure, CT and sonography were highly suggestive of compartment syndromes over the gluteal area. Gluteal compartment syndrome is an unusual phenomenon. Documented causes have included trauma, drug abuse accompanied by altered mental status and prolonged stasis.[10] Compartment syndrome is not always easy to identify in the early stage due to lack of clinical awareness, variability in the presenting complaints, and disturbed consciousness of patients during their initial visits. Thus, the differential diagnosis of painful swelling in the lower extremities presenting as deep vein thrombosis or post-traumatic sciatic nerve palsy should include compartment syndrome.[11] Unfamiliarity with this syndrome may delay necessary surgical treatment and lead to permanent disability. The most common cause of gluteal compartment syndrome is substance abuse accompanied by prolonged immobilization.[10–13] Other causes such as contusion, strenuous exercise and iatrogenic complication have also been reported.[14–17] A comparison of treatment effects from a review of the literature is shown in Table 3.

For plexus injury, conservative treatment is usually undertaken in the first few weeks. Surgical exploration is indicated in the absence of clinical or electromyographic recovery, and should be performed within 6 months.[18] Rehabilitation programs include electrical stimulation applied over weak muscles such as the anterior tibialis, gastrocnemius, and quadriceps, ambulation training with quadricane, strengthening exercises, and activities of daily living training. It was reported that percutaneous electrical stimulation could result in significant increase in muscle cross-sectional

Table 3. Review of literature for gluteal compartment syndrome

Authors	Etiology	Treatment	Outcome
Hynes et al (1994)[11]	Substance abuse	Conservative treatment	EMG/NCS showed persistent sensory loss and axonal degeneration, with regeneration 4 months later
Prynn et al (1994)[12]	Substance abuse	Fasciotomy	Return of normal sensation of the affected limb 3 days later, but then lost to follow-up
Bleicher et al (1997)[10]	Substance abuse (bilateral lower limbs were involved)	Fasciotomy	Weakness of abduction and external rotation, and walked without assistive device 1 week later
Klockgether et al (1997)[13]	Substance abuse	Fasciotomy	Atrophy of gluteus maximus and mild impaired sensation of light touch and pain in the affected leg 4 months later
Wise et al (1997)[15]	Exercise-induced (bilateral lower limbs were involved)	Fasciotomy	Walked with crutches 2 weeks later, returned to work as a salesman 1 month later, resumed weight lifting, running and competitive sports 2 months later, and complete recovery 6 months later
Krysa et al (2002)[16]	After posterior cruciate ligament surgery	Fasciotomy	Complete recovery (time not mentioned)
Roth et al (2002)[17]	After bone marrow biopsy	Fasciotomy	Foot drop persisted, near-normal thigh flexion and plantar flexion after 11 days
David et al (2005)[14]	Contusion injury (bilateral lower limbs were involved)	Fasciotomy	Trendelenburg gait persisted, sciatic nerve palsies resolved fully 15 months later

EMG/NCS = electromyography/nerve conduction study.

area, isokinetic peak torque, maximal isometric and dynamic strength, and motor performance skills.[19] Percutaneous electrical stimulation was also shown to prevent muscle atrophy after prolonged immobilization.[20] In this case that we have described, electrical stimulation and strengthening exercises were primarily focused on muscles for ambulation stabilization, which is mainly a functional concern. Strengthening exercises such as ankle pumping, quadriceps sets, and towel wringing were taught to the patient as exercises to be performed at home, 30 minutes each session, 3 sessions a day. The early intervention with rehabilitation prevented joint contracture and muscle atrophy in our patient.

In conclusion, the combination of brachial plexus and lumbosacral plexus injuries after prolonged compression is a rare but potentially debilitating phenomenon. The possibility of developing into gluteal compartment syndrome makes the condition even more difficult for the rehabilitative team to approach. Therefore, early diagnosis is crucial for prevention of local ischemia and further neurologic deterioration. Diagnosis based on clinical presentations, laboratory data, and EMG/NCS results should be kept in mind for early intervention and treatment.

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