

Case Report

Hitchhiker's Toe and Lumbar Disc Lesion: A Strange Coincidence in a Patient with Idiopathic Dystonia Treated with Botulinum Toxin Injection

Ayyoub Baqer¹, Jai Shanthini S², Wael Abdul Gawad³

Abstract

A 40 years old female with generalised dystonia and chronic low back pain was injected with botulinum toxin injection to extensor hallucis longus (EHL) for management of Hitchhiker's toe. The patient benefited functionally but later developed EHL and extensor digitorum (ED) weakness. Further workup revealed degenerative L4/5 disc lesion causing L5 root affection as the cause for the weakness. This coincidental occurrence of EHL and ED weakness due to L4/5 disc lesion in a patient with generalised dystonia, following botox injection to EHL for management of Hitchhiker's toe, has not been reported before.

Key words: Dystonia, Hitch hiker's toe, botulinum toxin injection.

Introduction:

Hitchhiker's toe has been defined as an apparent spontaneous extensor plantar response, without fanning of the toes. Other terminologies include striatal toe¹ or persistent extension of great toe or spontaneous Babinski sign or overactivity of the extensor hallucis longus (EHL). It occurs in patients suffering from stroke and dystonic syndromes. It causes painful spasms and gait disturbances including abnormal push-off and forward propulsion towards final stance². Botulinum toxin injection is effective in the management of Hitchhiker's toe seen in patients with dystonic syndromes. Dystonia could also result in early spinal degeneration³. In this report we present a patient with idiopathic dystonia who was treated with botulinum

toxin A injection to extensor hallucis longus muscle for the management of Hitchhiker's toe and was later found to have EHL and ED weakness due to degenerative lumbar disc lesion.

Case Report:

The patient was a 40 years old Caucasian female who suffered from generalised dystonia. She was born by caesarian section and was one of twins. She had completed secondary school and was employed in a clerical job. Her main complaint was painful spasms involving the trunk and lower limbs resulting in recurrent ankle injuries since childhood for which she had consulted neurologist for the first time at the age of 35 years. CT brain done twice was normal. Work up for secondary dystonia including serum ceruloplasmin was negative. She improved symptomatically with trihexyphenidyl (Artane) 2 mg twice daily. Subsequently the dystonic movements had decreased.

However she continued to have difficulty in walking due to bilateral persistent extension of great toes and her shoe uppers were getting torn frequently. She also complained of chronic low back pain of 5 years duration and spasms in both hands while writing suggestive of writer's cramps.

On examination, patient was found to have mild dysarthria; muscle power was normal in both upper limbs and in bilateral hips, knees and ankles. Dystonic movements were observed in both feet in the form of persistent extension of bilateral great toes and mild

Authors' affiliation:

¹ MD, FRCPC, Department of PMR

² MD, DNB, Department of PMR

³ MD, Department of PMR

Department of Physical Medicine & Rehabilitation, AI Farwaniya Hospital, Kuwait.

Cite as:

Ayyoub Baqer, Jai Shanthini S, Wael Abdul Gawad, Hitchhiker's Toe and Lumbar Disc Lesion a Strange Coincidence in a Patient with Idiopathic Dystonia Treated with Botulinum Toxin Injection, IJPMR, December 2015; Vol 26(4) :111-13

Correspondence:

Dr. Jai Shanthini Singaram, Department of Physical Medicine & Rehabilitation, AI Farwaniya Hospital, Kuwait.

Email: jaishanthini@gmail.com

Received on 03/05/2015, Accepted on, 20/11/2015

inversion of left foot which was exacerbated during walking. The great toe hyperextension was functionally disabling for the patient and hence it was decided to treat it with botulinum toxin injection. Injection botulinum toxin A was injected at therapeutic dose (Dysport 100 units) to bilateral extensor hallucis longus muscle after localising the muscle with ultrasonogram.

Patient was reviewed four weeks after the injection. We used the following scale for Shoe difficulties and pain : 0- absent 1- mild, 2- severe and 4 point scale for EHL overactivity as 0- no overactivity, 1- triggered by walking, 2- triggered by standing up, 3- triggered by any stimulation. The functional benefits after the injection in our patient were as follows:

On both sides, there was no more pain or damage to shoe upper. Pain and shoe difficulties scale was 2 preinjection and 0 post injection. On the right side, the EHL overactivity was still present but it was lesser - pre injection scale was 3 and post injection scale 1.

However on the left side, there was weakness of EHL as well as extensor digitorum (ED). She also complained of radiating pain to the left lower limb and exacerbation of low back pain since two weeks. There was tenderness in the lumbar spines L4 and L5 with painful restriction of lumbar flexion. On left side, there was weakness of extensors of all toes and diminished sensation in L5 dermatome. MRI confirmed L4/5 disc bulge with caudal migration and compression of bilateral L5 roots (Figs 1 & 2). Nerve conduction study of lower limbs was normal and patient did not agree for needle EMG. Neurosurgeon advised surgery for the L4/5 disc lesion but patient opted for conservative treatment. She was started on celecoxib 200 mg per day and pregabalin 75 mg twice a day and prescribed exercise programme and educated in back care; she reported decrease in pain with this treatment.

Discussion:

Dystonia:

Dystonia is defined as a movement disorder characterised by sustained or intermittent muscle contractions causing abnormal, often repetitive movements, postures, or both. Dystonic movements are typically patterned, twisting, and may be tremulous. Dystonia is often initiated or worsened by voluntary action and associated with overflow muscle activation⁴. The presentation is often variable. Our patient had generalised dystonia affecting different parts of the body variably including mild dysarthria, painful contractions of trunkal muscles, foot inversion during walking and sustained contraction of the extensor hallucis longus muscle.

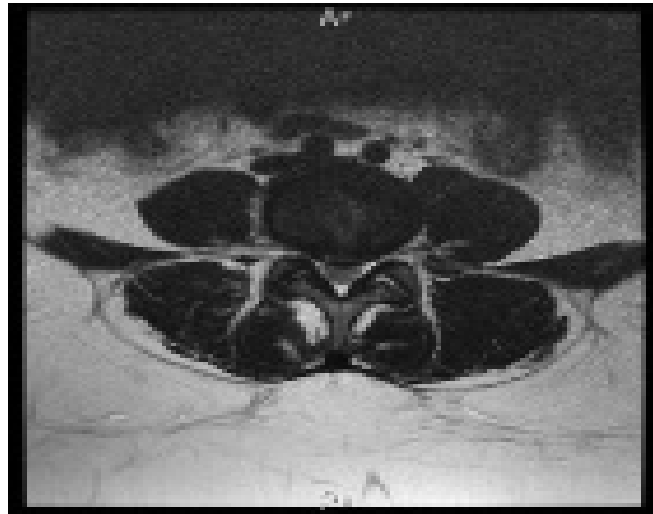


Fig 1: MRI Lumbar Spine, Showing L 4/5 Disc Bulge with Bilateral L5 Root Compression (Sagittal Weighted T2 images)



Fig 2: MRI Lumbar Spine, Showing L 4/5 Disc Bulge with Bilateral L5 Root Compression (Axial T2 Weighted Images)

Disabilities due to Hitchhiker's toe:

Hitchhiker's toe is disabling because of pain, shoe difficulties and abnormal posture. Complications include instability and falls. Our patient suffered from recurrent ankle sprains. The deformity was so severe that it was resulting in damage to her shoes.

Management of Hitchhiker's toe:

Surgery was the only effective strategy to manage EHL hyperextension before the introduction of botulinum toxin. Recent studies have proved that botulinum toxin

A is effective in the management of Hitchhiker's toe⁵. Yelnik *et al*⁶ described a case series of 11 patients with hemiplegia and overactivity of EHL; with post botulinum toxin injection, EHL overactivity disappeared and there was good subjective improvement of pain and shoe difficulties. The authors also described the scale to assess outcome after treatment which we have applied in this report. In our patient with generalised idiopathic dystonia, botulinum toxin A injection to EHL was effective in the management of Hitchhiker toe ; there was decrease in pain and shoe difficulties improvement in the severity of the overactive EHL and improvement of gait.

Weakness following botulinum toxin injection:

Botulinum toxin causes weakness in the muscle by neuromuscular blockade. After injection the toxin diffuses into the muscle and adjacent tissues. Adjacent muscle weakness can occur as side effect if high volumes are used⁷. Generalised weakness due to toxin spreading in blood is very rare. Our patient developed weakness in extensor hallucis longus and extensor digitorum on the left side. Ankle dorsiflexion and plantar flexion were normal. However the history of acute episode of low back pain with radiation to left lower limb, presence of sensory impairment in L5 and MRI finding of L4/5 disc lesion with L5 root compression confirmed that the weakness was the result of the disc lesion and not a side effect of botulinum toxin.

Spinal lesions and dystonia:

There are reports about cervical spine degenerative changes in patients with movement disorders. Hirose and Kadoya⁸ proposed that chronic involuntary movements seen in dystonia and athetosis contribute to the early degenerative changes in cervical spine. The mean age in their review of 251 cases was 33 years. Waterston *et al*⁹ reported two cases of idiopathic dystonia with cervical spondylotic myelopathy. A search in Pubmed did not find any report on a causal relation between dystonia of trunk muscles and lumbar disc lesion. Our patient was 40 years old with history of dystonic movements since childhood ; the movements predominantly involved the trunk and lower limb muscles. Her back pain had started at the age of 35 years . It can be hypothesised that the repetitive dystonic movements could have been a contributory factor for the early development of lumbar disc lesion with neurological deficit.

Hitchhiker's toe and lumbar disc lesion:

L4/5 disc lesion can present with Hitchhiker's toe. Blunt *et al*¹⁰ reported a patient with dystonic contraction of bilateral great toe extensors and MRI

findings of marked lumbar canal stenosis and bilateral L5 and S1 roots compression. The dystonic movements were ameliorated after spinal decompression. They hypothesised that lumbar disc lesion can produce foot dystonia. In our patient the dystonic movements had preceded the low back pain by several decades and the foot dystonia was part of the picture of generalised dystonia.

Conclusions:

Botulinum toxin A injection is effective in the management of dystonic Hitchhiker's toe. When unexplained lower limb weakness occurs in lower limb after botulinum toxin injection in dystonic patient, a careful search for lumbar disc lesion should be made.

References:

1. Winkler AS, Reuter I, Harwood G, Chaudhuri KR. The frequency and significance of 'striatal toe' in parkinsonism. *Parkinsonism Relat Disord* 2002; **9**: 97–101.
2. Bensoussan L, Viton JM, Barotsis N, Delarque A. Evaluation of patients with gait abnormalities in Physical Medicine and Rehabilitation settings. *J Rehabil Med* 2008; **40**: 497–507.
3. Jameson R. Cervical myelopathy in athetoid and dystonic cerebral palsy: retrospective study and literature review; *Eur Spine J* 2010 ;**19**:706–12.
4. Albanese A, Bhatia K, Bressman SB, *et al*. Phenomenology and classification of dystonia: a consensus update. *Mov Disord* 2013 ; **28**: 863–73.
5. Gaber *et al*. Botulinum toxin in the management of hitchhiker's toe. *Neurorehabilitation* 2011; **28**:395 -9.
6. Yelnik AP, Colle FM, Bonan IV, Lamotte DR. Disabling overactivity of the extensor hallucis longus after stroke: clinical expression and efficacy of botulinum toxin type A. *Arch Phys Med Rehabil* 2003;**84**. Jan 2003.
7. Manchau A, Bhatia KP. Uses of botulinum toxin injection in medicine today. *BMJ* 2000; **320**, Jan 161-5.
8. Hirose G, Kadoya S. Cervical spondylotic myelopathy in patients with athetoid dystonic cerebral palsy : clinical evaluation and surgical treatment. *Neurol Neurosurg Psychiatry* 1984; **47**: 775 -80.
9. Waterston JA, Swash M, Watkins ES. Idiopathic dystonia and cervical spondylotic Myelopathy. *Neurol, Neurosurg Psychiatry* 1989; **52**: 1424-6.
10. Blunt SB, Richards PG, Khalil N. Foot dystonia and lumbar canal stenosis. *Mov Disord* 1996; **11**:723-5.