Runner's Dystonia

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ABSTRACT

OBJECTIVE: To characterize a unique form of task-specific dystonia involving proximal lower limbs during long-distance running. **BACKGROUND:** Adult-onset focal dystonia in upper limbs is well characterized whereas such dystonia has been rarely reported in lower limbs, especially in proximal parts. When such focal dystonia occurs in an athlete it is often wrongly attributed to an orthopedic disorder. With the possible exception of "golfer's yip", adult-onset, task-specific focal dystonia involving the lower limb is quite rare and runner's dystonia has not been previously reported. **METHODS:** A database of 622 patients with focal limb dystonia was reviewed for evidence of focal task-specific dystonia precipitated by running. In addition to examination, the patients were videotaped while sitting, walking and running. **RESULT:** We present five cases, three female and two male with mean age of 44.6±10.4 years, mean age at onset of 37.4±10.3 years, and mean duration of symptoms for 7.2±4.4 years, who initially noted dystonia of one leg during long-distance running. The clinical features of dystonia in these long-distance runners overlap with those of more recognizable forms of focal dystonia including relief with sensory "tricks". They also share features with paroxysmal dyskinesia, as carbamazepine markedly ameliorated the symptoms at least in one patient. One patient benefited from oral anticholinergic, one from levodopa, and three patients benefited from repeat botulinum toxin injections. Our patients differed from the typical childhood-onset leg dystonia, such as the DYT1 dystonia, in that there was no family history of dystonia and the leg dystonia in our patients remained focal and did not spread to other body parts. Two of our patients had prior injury to the affected leg within one year prior to the onset of the dystonia, raising the possibility of peripherally-induced dystonia. **CONCLUSION:** We describe five patients with runner's dystonia and draw attention to this unique and disabling, adult onset focal dystonia involving predominantly proximal lower limbs. When recognized early, it may be treated effectively with anticholinergic drugs, anticonvulsants, levodopa or botulinum toxin injections.

INTRODUCTION

1. Dystonia is characterized by involuntary, sustained muscle contractions causing twisting movements or abnormal postures [1].
2. Focal dystonia affects only a single isolated body part with symptoms varying from action or even task-specific dystonia to a fixed abnormal posture [2]. In patients with adult-onset dystonia, focal dystonia manifested as blepharospasm, oromandibular dystonia, and torticollis, is most common[3].
3. With the possible exception of "golfer's yip" which has been receiving increasing attention [4,5], adult-onset, focal dystonia in lower limbs in athletes has rarely been reported.
4. Dystonia in long-distance runners has never been described in the literature. Here we report five cases of adult-onset focal dystonia involving mainly proximal lower limbs occurring initially during long-distance running.

METHODS

A database of 622 patients with focal limb dystonia was reviewed for evidence of focal task-specific dystonia precipitated by running. In addition to examination, the patients were videotaped while sitting, walking and running.

RESULTS

TABLE 1. Demographic and clinical characteristics of all patients

Case	#1	#2	#3	#4	#5	Mean
Sex	F	F	F	M	M	
Age (years)	40	49	58	30	46	44.6 ± 10.4
Age at Onset	37	40	46	20	44	37.4 ± 10.3
Duration (years	3	9	12	10	2	7.2 ± 4.4
Prior injury	+	+	-	-	-	
Family History	-	-	-	-	-	
Limb involved	L	L	L	L	R	
Initial presentation	foot flexion	Knee buckling	Foot inversion 20 minutes after running	Knee extension and foot inversion while running	Foot flexion during the last quarter-mile of his ninth marathon running in 4 years	
Clinical manifestation	Left knee extension	Left knee extension and left hip flexion	Left knee extension, left foot eversion induced by running	Left knee extension, left foot inversion	Right foot plantar flexion	
Trigger	walking on tip toes	squatting	Biked more than 20 minutes	Walking upstairs	Running with shoes on	
Associated dystonic tremor	-	+	+	-	-	
Null point	-	+ With knee flexion and leg abduction in a supine position	+ with leg flexion and abduction	-	-	
Sensorimotor trick	+ Walking backwards, with partially flexed knees, on heel	+ Touching left hip or sitting with back touching the chair, walking on heels	-	+ Walking backwards	-	
EMG/NCV	Normal	Normal	Normal	Normal	Normal	
MRI (brain and spine)	Normal	Normal	Normal	Normal	Normal	
DYT1 gene	-	NA	NA	-	-	
Effective treatment	Carbamazepine	Botulinum Toxin	Levodopa	Trihexyphenidyl	Botulinum Toxin	
Response	marked	marked	marked	moderate	mild	

F: female, M: male, AAO: age at onset, L: left, R: right, NA: not available

DISCUSSION

1. All five patients, were avid, long-distance runners whose athletic performance became markedly impaired as a result of action-induced dystonia of lower limbs.
2. The presence of sensory or motor tricks, also supports the diagnosis of dystonia. Tremor, a common feature of focal dystonia[6,7], was presented in cases 2 and 3. Both patients also exhibited another characteristic feature of dystonic tremor, namely a "null point", a position in which the tremor subsides or resolves.
3. Adult onset foot or toe dystonia is quite rare[8-10] and proximal leg involvement is even rarer and is usually misdiagnosed as an orthopedic or psychogenic disorder.
4. In contrast to primary dystonia starting in childhood, typified by DYT1 dystonia[11], the leg dystonia in our patients remained focal and did not spread to other body parts.
5. Two of our patients had prior injury within one year of the onset and the symptoms occurred anatomically related to the site of injury, raising the possibility of peripherally-induced dystonia .
6. Immobilization, including casting, has been previously suggested as a precipitant of peripherally-induced dystonia[13]. Since patients with runner's dystonia may be misdiagnosed as having an orthopedic problem and casting may be recommended, the reported cases of casting-induced dystonia should caution against such practice.
7. All patients presented with similar clinical phenotype but responded to different treatments. Two patients (case 2 and 5) who did not have satisfactory response to oral medications responded to botulinum toxin injections.
8. One patient (case 3) responded well to levodopa, similar to patients with Parkinson's disease and levodopa-responsive dystonia[14]. However, the late age at onset, the focal nature, and lack of diurnal fluctuation argued against the diagnosis of levodopa-responsive dystonia.

CONCLUSION

In summary, we describe 5 patients with runner's dystonia and draw attention to this disabling, adult onset focal dystonia involving predominantly lower limbs. Since all patients were initially misdiagnosed we suspect that this disorder is more common than suggested by this small series. When recognized early, potentially risky treatments such as immobilization may be avoided, and anticholinergic drugs, anticonvulsants, levodopa or botulinum toxin injections may be tried to relieve the otherwise disabling spasm and abnormal postures. Similar to other task-specific dystonias, runner's dystonia does not appear to progress or spread to other body parts, although it often markedly curtails the athletic performance.

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