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Focal task-specific lower extremity dystonia associated with intense repetitive exercise: A case series

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ABSTRACT

Background: Focal task-specific dystonia of the lower extremity associated with intense repetitive exercise has recently been recognized. The clinical course, treatment response and prognosis remain poorly understood.

Methods: Individuals with lower extremity task-specific dystonia evaluated at UCSF's Movement Disorders Center (2004–2012) were eligible for this descriptive case study series if he/she had a history of strenuous and prolonged exercise involving the lower extremity and had no abnormal neurological or medical conditions to explain the involuntary movements. Data was gathered from the medical history and a self-report questionnaire. The findings were compared to 14 cases previously reported in the literature.

Results: Seven cases (4M/3F) were identified with a diverse set of exercise triggers (cycling, hiking, long-distance running, drumming). The mean age of symptom onset was 53.7 ± 6.1 years. The median symptom duration prior to diagnosis was 4 (9.5) years. Several patients underwent unnecessary procedures prior to being appropriately diagnosed. Over a median of 2 (3.5) years, signs and symptoms progressed to impair walking. Seven patients had improvement in gait with treatment (e.g. botulinum toxin injections, benzodiazepines, physical therapy, bracing, body weight supported gait training and/or functional electrical stimulation of the peroneal nerve) and six returned to a reduced intensity exercise routine.

Conclusions: Isolated lower extremity dystonia associated with strenuous, repetitive exercise is relatively uncommon, but disabling and challenging to treat. The pathophysiology may be similar to task-specific focal dystonias of the upper limb. Prompt recognition of leg dystonia associated with extreme exercise could minimize unnecessary testing and procedures, and facilitate earlier treatment.

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1. Introduction

Dystonia is a central nervous system disorder characterized by disabling, involuntary, sustained or intermittent muscle contractions [1,2]. These patterned contractions lead to twisting movements and abnormal postures that can be further categorized by the distribution of symptoms. Dystonia may affect a specific body region (focal dystonia) or multiple body regions (segmental, multifocal, hemibody or generalized dystonia), performance of a specific activity (task-specific dystonia), or spread to similar tasks.

Task-specific focal hand dystonia following intensive, repetitive use of the upper extremity has been described in musicians [3], writers [4], computer keyboard users [5], and athletes (e.g. table tennis players [6] and golfers [7]). Isolated focal task-specific dystonia of the lower extremity associated with strenuous repetitive exercise has recently been reported in the literature [8–13]. With the objective to increase knowledge and facilitate earlier diagnosis and treatment of this condition, we describe seven cases of focal task-specific dystonia of the lower extremity associated with intense, repetitive exercise evaluated at our Movement Disorders Center. We also compare our patient cohort with previously published cases.

2. Methods

Individuals with lower extremity task-specific focal dystonia who were evaluated at our Movement Disorders Center between 2004 and 2012 were eligible for this case series if they had a history of strenuous and prolonged exercise involving the lower extremities prior to symptom onset occurring for many hours a week for

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Table 1
Case descriptions.

Patient number	#1	#2[a]	#3	#4	#5[b]	#6	#7
Sex	M	M	F	M	F	F	M
Age (years)	57	69	68	61	69	50	75
Level of activity prior to dystonia (average)	Running 30 miles per week for 20 years, then switched to biking 200 miles per week for one year	Running 80 miles/week for 22 years	Running 25 miles/week for 45 years, then switched to biking 55 miles per week, walking 10 miles per week, and kickboxing 1 h per week for 3 years	Running 30 miles per week for 40 years	Walking and hiking multiple times per week for at least 5 years	Running 8 miles per week for 9 years, then 15 miles per week for 8 years. Escalated running one month prior to symptom onset to 60 miles per week	Biking 175 miles/week for 15 years, running 10 miles/week for 15 years, drumming 30 h/week for 30 years
Age at onset (years)	52	54	64	54	58	49	45
Duration (years)	5	15	4	7	11	1	30
Years before diagnosis	3	4	2.5	4	11	0.25	13
Alternative diagnoses given	Knee osteoarthritis	Baker's cyst, advanced age, SCA	PD, primary lateral sclerosis	Right acetabular labral tear and impingement syndrome	Foot drop, neuropathy, radiculopathy, myelopathy	Tarsal tunnel syndrome, Sinus Tarsi syndrome	Performance anxiety (self-diagnosed)
Unnecessary procedures or surgeries?	Right knee cortisone injections and right knee arthroscopic surgery to remove hardware (screw) from a prior ACL repair	No	No	Right hip cortisone and platelet-rich-plasma injections, offered surgery for right impingement syndrome	No	Marcaine injections for sinus tarsi syndrome, surgery for sinus tarsi syndrome recommended	No
Initial exercise trigger(s)	Running, biking	Running, biking	Running, biking, kickboxing, walking	Running	Hiking, walking	Running	Running, biking, drumming
Prior injury	No	No	No	No	No	No	No
Painful dystonia?	No	No	No	Yes	No	No	No
Family history	Yes	No	No	No	No	No	No
Clinical manifestation	Right plantarflexion, foot inversion, medial leg rotation and adduction, hip flexion and adduction while ambulating	Right dorsiflexion, knee hyperextension, hip abduction and toe extension while ambulating; right toe extension at rest	Right foot inversion, plantarflexion and toe curling while ambulating	Right hip abduction, foot eversion and plantarflexion, as well as left toe curling and plantarflexion while ambulating	Left plantarflexion and foot inversion while ambulating	Left toe curling while ambulating	Right plantarflexion while ambulating
Geste antagoniste	No	No	No	No	No	No	Using heel to drum while placing toes on a higher surface
Progression to affect walking (years)	2	3	1	4	0.5	0.08	20
Spread to other body parts	No	No	No	Contralateral leg 4 years after onset	No	No	No
MRI brain	Very mild generalized atrophy	Right frontal meningioma	Normal	Mild white matter ischemic disease	Unknown	Normal	Mild white matter ischemic disease
Genetic testing	Not done	SCA 1,2,3,6,7 negative DRPLA negative	Not done	Not done	Unknown	Not done	Not done
Effective treatment	Botulinum toxin Clonazepam	Physical therapy Body support treadmill	Physical therapy FES: peroneal nerve Botulinum toxin	AFO brace Botulinum toxin Lorazepam Physical therapy FES: peroneal nerve	AFO brace Physical therapy Botulinum toxin	Botulinum toxin	AFO brace Botulinum toxin Physical therapy FES: peroneal nerve
Return to exercise	Yes	Yes	Yes	Yes	Yes	No	Yes

AFO – ankle-foot orthotic, FES – functional electrical stimulation; LLE – left lower extremity; PD – Parkinson's disease; SCA - spinocerebellar ataxia.

[a] See Video I.

[b] Lost to follow-up.