SHANE MCCLINTON, PT, DPT, OCS, FAAOMPT, CSCS[1] • **BRYAN HEIDERSCHEIT,** PT, PhD[2]

Diagnosis of Primary Task-Specific Lower Extremity Dystonia in a Runner

Dystonia is a dynamic neurological syndrome associated with involuntary and sustained muscle contraction that causes abnormal postures and movement.[5,13] One of the key distinguishing characteristics of dystonia is the dynamic nature of its presentation.[5] Examination findings may appear inconsistent because specific (focal) or general movement/postural abnormalities can present differently depending on posture, position, or the intended task.

The 2 types of dystonia, based on clinical and genetic epidemiological studies, are primary and secondary. Most documented cases of dystonia are of primary etiology,[27] where there is no evidence in the history, examination, or laboratory studies (including structural abnormalities in the central nervous system) that can identify a cause. Conversely, secondary dystonia is believed to result from physical or neurological trauma, medicinal or environmental toxicity, or psychogenic factors.[5] Furthermore, primary and secondary dystonia are categorized based on the extent of the body regions affected, to include focal (involvement of only 1 body region), segmental (involvement of continuous body regions), or general (involvement of multiple body regions).[5] Task-specific dystonia is a form of focal dystonia that results in abnormal movements and postures exclusively within the context of a specific task. Most reported cases of task-specific focal dystonia involve the upper extremity, including syndromes such as writer's cramp, musician's dystonia, and golfer's yips.[39] In each of these syndromes, the altered movement pattern is specific to a task that has been performed repetitively and often plays a substantial role in the affected individual's livelihood. The distinguishing feature of runner's dystonia from other task-specific dystonias is that the individual first develops symptoms during running.[41]

Although the overall prevalence of runner's dystonia is unknown, isolated population estimates of primary focal dystonia in the United States are 295 per million persons.[27] Available data from the same population provide a relative appreciation of the prevalence of focal dystonia compared to more commonly known movement-related disorders such as Parkinson's disease (1569 per million), amyotrophic lateral sclerosis (64 per million), and convulsive disorders (6500 per million).[27] Outside of the United States, the prevalence of individuals with pri-

STUDY DESIGN: Resident's case problem.

BACKGROUND: A 56-year-old man was referred to physical therapy for analysis of unusual gait, first noticed 3 years previously when running. Prior to this evaluation, the patient had seen multiple orthopaedic, sports medicine, and neurological specialists while undergoing repeated and extensive testing. Ten months of testing and treatment, including conservative and surgical management, did not provide an explanation for the gait abnormality or result in improvement of the patient's condition.

DIAGNOSIS: The patient's physical examination was relatively unremarkable, considering the severity of the gait abnormality. Distinct abnormalities were apparent with computerized gait analysis and dynamic electromyography, and, when combined with the physical examination findings, led to a suspicion of the task-specific disorder of runner's dystonia. The patient was referred to a neurologist specializing in movement-related disorders, with a final confirmed diagnosis of primary task-specific dystonia with first onset during running (ie, runner's dystonia).

DISCUSSION: Idiopathic, task-specific dystonia of the lower extremity is documented as a very rare occurrence, yet increasing trends in running participation may result in a higher incidence of this condition. Improved awareness of runner's dystonia in the present case might have enhanced the clinical decision-making process and resulted in more timely and effective treatment solutions. Clinical examination findings, including computerized gait analysis and electromyography, in conjunction with imaging, blood, and genetic testing, can aid in the diagnosis of runner's dystonia.

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[1]Physical Therapist, Des Moines University Clinic, Des Moines, IA. [2]Associate Professor, Department of Orthopedics and Rehabilitation, University of Wisconsin-Madison, Madison, WI. Following evaluation and prior to presentation of this case, approval was obtained from the Des Moines University Institutional Review Board, and the individual in this case provided consent for presentation of his medical information and videos. Address correspondence to Dr Shane McClinton, Des Moines University Clinic, 3200 Grand Avenue, Des Moines, IA 50312. E-mail: shane.mcclinton@dmu.edu

mary focal dystonia who presented to outpatient clinics ranges between 61 and 137 per million persons.[9,11,16,26,31] Whereas primary focal dystonia is relatively rare, task-specific lower extremity dystonia is even less prevalent. Within a specialized movement-disorder clinic, only 4 (0.7%) of 579 individuals with primary focal dystonia had lower extremity dystonia,[21] and none of these individuals presented with the specific movement pattern described in this case. With growing numbers of runners, including participants in high-mileage events (marathons and ultra-marathons), a higher prevalence of lower extremity dystonia, including runner's dystonia, is likely. Epidemiological investigation to accurately define the prevalence of lower extremity dystonia is hindered by the perception that dystonia is rare and the heterogeneous presentation of the condition.[9] A lack of current epidemiological data specific to runner's dystonia contributes to a lack of awareness and consideration in clinical diagnostic decisions. This may result in missed diagnosis and inappropriate or ineffective treatment recommendations.

In this case report, we will describe the circuitous diagnostic path and initial treatment of an individual who was ultimately determined to have runner's dystonia. The purpose is to increase awareness of runner's dystonia such that it may be more readily recognized and managed efficiently.

DIAGNOSIS

History

A 56-year-old man was referred to physical therapy (PT2) by his family practice physician for 3-D motion analysis with a diagnosis of gait dysfunction. The patient's gait dysfunction was first noted almost 3 years prior when the left lower extremity became clumsy and awkward during his usual running routine. The patient was a consistent runner and had logged approximately 121,000 km (approximately 75,000 mi) throughout his lifetime. After initial onset, the symptoms progressed over the following 2 years until he could no longer run, followed by similar progressive decline in walking. He first presented to his primary care physician, describing the left-sided awkwardness during walking and running without pain and without further problems in other daily or recreational tasks. The dysfunction during walking involved minimal deviation for the first few steps, with progression to excessive left knee flexion at terminal swing that persisted through the following left midstance phase, resulting in reduced stride length (**ONLINE VIDEO**). His gait pattern would continue to degrade with walking continuously for slightly more than 1 minute, when he would need to stop for fear of falling. After only a few minutes of rest, he could walk again with minimal deviation until his gait pattern progressively deteriorated. The patient relayed that the only factor that improved his gait was walking on the beach, as observed while he was on vacation, and applying pressure with his hand to his hip.

Prior to the initial evaluation by PT2, the patient underwent multiple and repeated orthopaedic, cardiovascular, and neurological diagnostic testing, involving evaluation by 3 different physician orthopaedic specialists, a sports medicine physician, a neurologist, and another physical therapist (**TABLE 1**). He had failed to respond to cortisone injection, manual therapy, exercise, and orthopaedic surgery directed primarily at the hip on the involved side.

Examination

Review of the patient's medical history and review of systems was unremarkable, except for perceived muscle weakness, difficulty walking, and recent left hip offset osteotomy performed to correct the gait dysfunction. Subjectively, he reported 0/10 pain during walking and at rest. The patient did not indicate any history of neurological or medical red flags, including headaches, visual disturbances, loss of consciousness or balance, or bowel, bladder, diet, or weight changes. His Lower Extremity Functional Scale (LEFS) score indicated a clinically significant decline in function since his visit with the first physical therapist (PT1) 10 months prior (from 51/80 to 40/80; 80 is no disability). At present, he did not report any disability (0/100; 0% is no disability) on the Modified Oswestry Low Back Pain Disability Questionnaire. The patient did not complete a questionnaire specific to psychosocial status, but interactions with the patient and family indicated that there was a significant amount of stress related to his work and a busy family schedule outside of work. The family reported that the patient often had difficulty getting away from work or relaxing during family vacations.

Physical examination included muscle strength, range of motion, movement coordination, functional movement, and neurological testing (**TABLE 2**). Functional movement testing following observation of gait included bilateral deep squat, single-leg squat, single-leg step-up/step-down, and isolated demonstration of each phase of the gait cycle from mid-swing through midstance (fractionation). The patient demonstrated sufficient strength, coordination, and isolated functional movement to ambulate normally. All basic neurological testing was negative, including cranial nerve screen, deep tendon reflexes, sensation, Babinski, Hoffmann, and ankle clonus. It was apparent that the dysfunction was specific to the task of walking and not explained by the historical and initial physical examination findings.

Three-dimensional motion analysis was conducted while the patient walked at his preferred speed of 1.07 m/s (normal, 1.32-1.33 m/s)[35] repeatedly across a 6-m laboratory walkway. Motion analysis included collection of joint kinematics and kinetics, as well as surface electromyography (EMG) of bilateral medial hamstrings, vastus lateralis, gastrocnemius, and tibialis anterior (**TABLE 2**, **FIGURES 1** and **2**). Two-dimensional video analysis was also conducted from the sagittal and

TABLE 1 Timeline of Patient Testing and Management Prior to Evaluation by the Second Physical Therapist (PT2)

Time, mo	Event	Testing/Intervention	Significant Findings/Results	Recommendations
0	Left leg first felt clumsy/awkward with running	...	...	...
22	Forced to stop running	...	...	...
22.5	PCP consult	Physical exam	Observational gait abnormalities	Referral to Neuro1
22.75	Neuro1 consult	Neurological exam; radiographs: B hips and knees	No abnormalities to explain gait disturbance; no bony abnormality	None
23.25	Ortho1 consult	Physical exam	Observational gait abnormalities	Referral to Ortho2
23.5	SportMed1 consult	Physical exam	Observational gait abnormalities; suspected dynamic leg weakness	Referral to PT1
23.75	PT1 consult	Physical exam; 2-D video gait analysis	Positive Ober test L; L hip abductor MMT 4+/5; L hip IR ROM, 13° (R, 20°); 30° knee flexion at L heel strike	MRI of the hip; PT 2 times per wk for 4 wk
24	SportMed1 follow-up	Inguinal hernia examination	Negative	MRA of the hip
24.03	Rad1 consult	MRA L hip	L hip labral tear	...
24.25	Vascular consult	Vascular study	Normal	...
25	Ortho2 consult	Radiographs: B hips and pelvis	B cam–type femoral acetabular impingement, lateral femoral head prominences, decreased femoral head/neck offset; B early DJD, R acetabular rim fracture	Referral to Ortho3; continue PT; radiographs and MRI of lumbar spine
25 to 26	PT1 appointments	Manual therapy, therapeutic exercise, and gait training	No change in gait abnormality	...
26.3	Rad2 consult	Radiographs and MRI (with and without contrast) of lumbar spine	Mild disc space narrowing at L5-S1; normal MRI	...
26.5	Ortho3 consult	Physical exam; review imaging tests	No new findings	L hip cortisone injection; NCS/EMG study; surgery pending outcome of above
26.75	Ana1 consult	L hip cortisone injection	No improvement of gait	...
28.25	PMR1 consult	Static NCS and EMG	Normal studies	...
29.5	Surgery by Ortho3	L hip offset osteotomy and debridement	...	...
32	Follow-up by Ortho3	Repeat radiographs	Normal healing from osteotomy procedure; no improvement of gait	3-D motion analysis
32.25	PCP follow-up	...	...	3-D motion analysis with PT2
33	PT2 consult	3-D kinematic/kinetic motion analysis with dynamic EMG; physical examination	See **TABLE 2**	Referral to Neuro2
34	Neuro2 consult	Blood work; brain scan	Idiopathic task-specific dystonia	Botox injections; Parkinson's medication

Abbreviations: Ana, anesthesiologist; B, bilateral; DJD, degenerative joint disease; EMG, electromyography; IR, internal rotation; L, left; MMT, manual muscle test; MRA, magnetic resonance arthrography; MRI, magnetic resonance imaging; NCS, nerve conduction velocity study; Neuro, neurological physician specialist; Ortho, orthopaedic surgeon or physician specialist; PCP, primary care physician; PMR, physical medicine and rehabilitation physician specialist; PT, physical therapist; R, right; Rad, radiologist; ROM, range of motion; SportMed, sports medicine physician specialist.

frontal plane perspectives (**ONLINE VIDEO**).

Diagnosis

Based on the results of prior diagnostic testing (essentially normal radiographs, magnetic resonance imaging, and EMG/nerve conduction velocity study), prior treatment (lack of response to orthopaedic-based physical therapy and to hip offset osteotomy), and absence of pain, the likelihood of a musculoskeletal (femoroacetabular impingement, acetabular labral derangement) or peripheral neurodynamic (unilateral foraminal stenosis, lumbar plexopathy, lumbar radiculopathy) condition was considered to be very low. Given the findings from the physical examination, computerized

TABLE 2 Examination Findings of the Second Physical Therapist (PT2)

Tests	Significant Findings
3-D kinematic/kinetic motion analysis	Increased left knee flexion and ankle dorsiflexion at terminal swing through midstance phase, followed by increased right hip flexion, knee flexion, and ankle dorsiflexion (**FIGURE 1**) with elevated right knee extensor moment from midstance through preswing phase. Increased left hip flexion throughout swing phase and initial contact (**FIGURE 1**). Increased left ankle plantar flexion at terminal stance phase in compensation for the vertical depression of the pelvis due to findings described above (**FIGURE 1**). Decreased left step length and stance phase duration
Dynamic surface EMG	Early activation of hamstrings and absence of quadriceps activation during terminal swing phase on the left (**FIGURE 2**). Increased activation of left medial hamstrings, left gastrocnemius, and right vastus lateralis during stance phase, with increased right medial hamstrings during the last 20% of stance (**FIGURE 2**)
Lower Extremity Functional Scale	40/80 (80 is normal function)
Modified Oswestry Low Back Pain Disability Questionnaire	0% (0% is no disability)
Lower extremity strength (right, left)*	
Hip	
Flexion	5/5, 4/5
Extension	5/5, 5/5
Abduction	5/5, 4/5
Knee	
Flexion	5/5, 4+/5
Extension	5/5, 5/5
Ankle	
Dorsiflexion	5/5, 5/5
Plantar flexion	5/5, 5/5
Cranial nerve testing	Negative screening for cranial nerves I to XII[15]
Fractionation	Patient able to demonstrate isolated and segmental control of the thigh, lower leg, and foot, including reproduction of normal preswing and stance phase movements on the left when performed outside of gait
Flexibility/range of motion	Sufficient lower extremity flexibility and range of motion to achieve range of motion required during gait. No side-to-side differences appreciated
LMN screen	Ankle and knee deep tendon reflexes 2+ bilaterally. Normal light-touch sensation bilateral lower extremity dermatomes
UMN screen	Negative Babinski sign bilaterally. Negative Hoffmann sign bilaterally. No ankle clonus bilaterally. Normal heel-to-shin coordination bilaterally. Negative Romberg test

Abbreviations: EMG, electromyography; LMN, lower motor neuron; UMN, upper motor neuron.
**Assessed via the manual muscle test. Patient was able to perform single-leg squat bilaterally without deviation and with symmetry between the 2 lower extremities.*

gait analysis, and EMG, a neurological condition of the central nervous system was suspected, and PT2 consulted another physical therapist with expertise in gait analysis and motor control (PT3). PT3 recommended that the patient's presentation be compared to published case studies of individuals with runner's dystonia.[19,41] The patient had similar features to these cases, including (1) task specificity of the movement dysfunction, (2) long history of running with first onset of symptoms during running, (3) inconsistent orthopaedic examination (muscle performance, coordination, joint mobility, and fractionation testing not consistent with the observed gait pattern), (4) negative basic neurological examination, and (5) absence of pain. In addition, dynamic EMG demonstrated sustained and excessive muscular activation patterns common to task-specific dystonia.[5,39] The similarities evident in these case studies and the patient's presentation resulted in a high suspicion of runner's dystonia and prompted PT2 to refer the patient to a physician neurologist who specialized in movement-related disorders (Neuro2).

One month later, the patient was evaluated by Neuro2. Upon the initial examination, 3 diagnoses were considered: (1) an autoimmune glutamic acid decarboxylase antibody–associated abnormality (stiff leg syndrome), (2) dystonia, and (3) Parkinsonism. The following tests were performed to assist in the diagnosis: magnetic resonance imaging of the brain, serum ceruloplasmin, glutamic acid decarboxylase antibody testing, and genetic testing (particularly to identify presence of the dystonia 1, or DYT1, gene). In addition, a short trial of carbidopa-levodopa, typically used for Parkinson's disease, was prescribed to determine if the patient would be responsive to this medication. Even though the patient demonstrated no other signs or symptoms of Parkinson's disease, certain forms of dystonia have been responsive to Parkinsonian medication (dopa-responsive dystonia).[28] Based on the negative findings of the tests listed above and an unsuccessful trial of levodopa, the patient was diagnosed with primary task-specific dystonia (ie, runner's dystonia).

Treatment

Once the diagnosis of runner's dystonia was made, the patient was prescribed clonazepam, of which the patient subjectively reported improvement in walking duration with less perceived need of

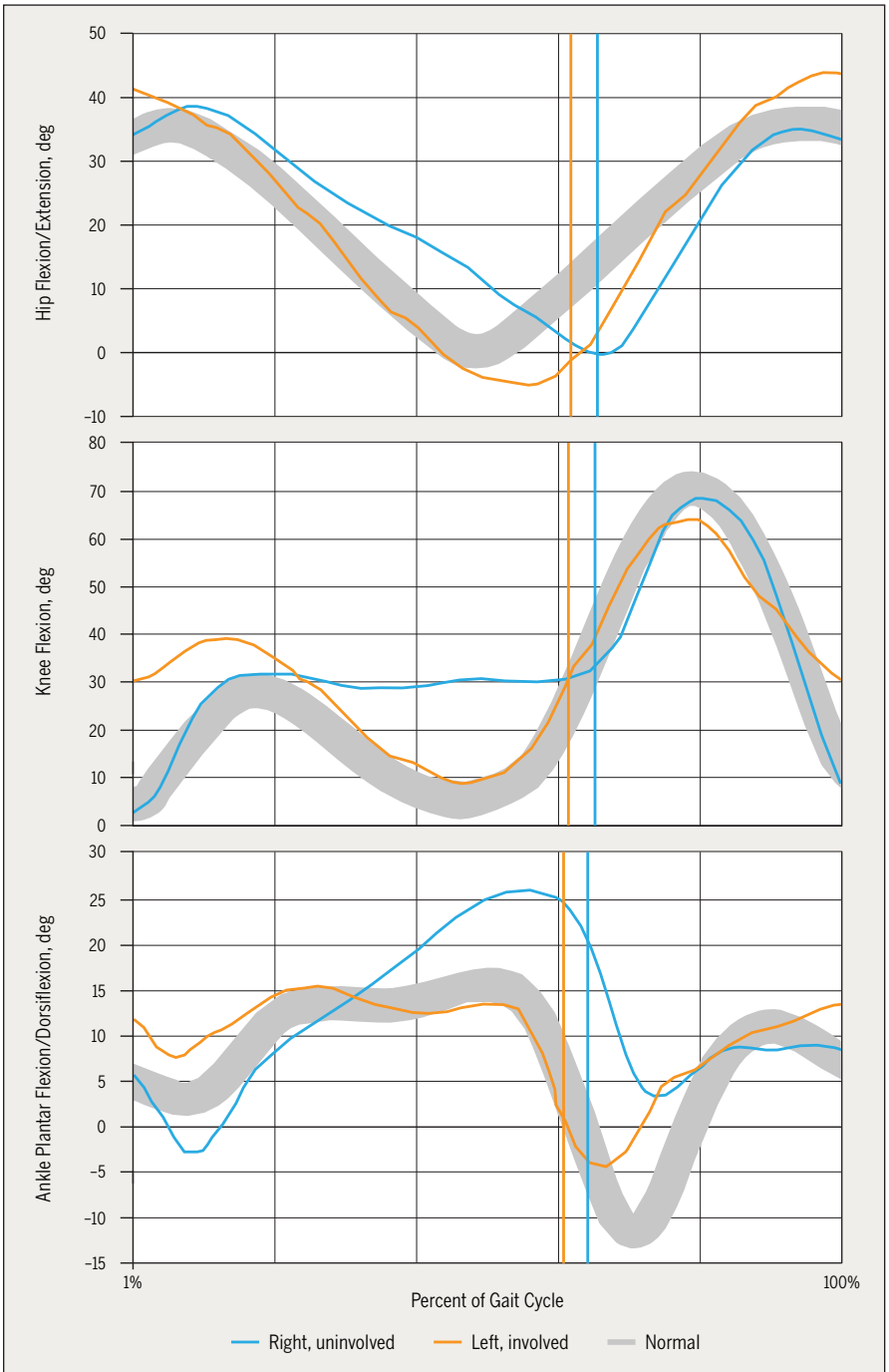

FIGURE 1. Kinematic impairments are present in the left hip, knee, and ankle primarily during terminal swing through midstance, and compensatory right-side impairments during midstance through preswing (**TABLE 2**). Positive values represent flexion or dorsiflexion during walking. The gait cycle is represented by initial foot contact (1%) and the point just before initial foot contact of the subsequent stride (100%). The vertical orange and blue lines depict toe-off for the left and right sides, respectively. The shaded gray region represents normal kinematic patterns during walking.[35]

walking aids. Six weeks following the diagnosis, the patient underwent a series of 3 botulinum toxin (Botox) injections 3 months apart in progressive doses (50, 100, and 150 U) to the left hamstring, with which he reported no additional improvement. Eleven months after the last Botox injection, the patient returned to PT2 for further intervention for the first time following confirmation of the primary task-specific dystonia diagnosis. The physical examination test and measures were repeated and remained similar to those of the first evaluation of the patient (**TABLE 2**). The only change was the LEFS score, which increased from 40/80 to 55/80. The patient attributed this improvement to the clonazepam.

The patient was questioned about potential movement or sensory tricks that could reduce the dystonic movement pattern. Identified tricks included minimal patient-perceived improvement with barefoot sand walking, a unilateral skipping pattern (where the hop only occurs on the uninvolved side), and manual pressure applied to the involved hip. Additional gait variations, including walking backward, sidestepping, body-weight-supported treadmill walking, and shortened step length, were attempted but did not alter the dystonic gait pattern. At this visit, sensorimotor testing was also performed, based on the category of tests used in a recent clinical trial involving individuals with focal hand dystonia[6] and as recommended in the assessment of sensorimotor function.[30] These tests were adapted for the foot to allow for application to this case (**TABLE 3**).

Due to a lack of evidence on management of lower extremity dystonia, the treatment plan for this patient was based on the management of task-specific dystonia of the upper extremity[6] and management of similar disorders that involve cortical reorganization, such as chronic regional pain syndrome or phantom limb pain.[24,25] The components of the program included physical fitness, brain fitness, and learning-based sensorimotor training, including graded motor imagery.[6]

The patient required little guidance on physical fitness because he already exer-

cised 3 to 5 times per week, including aerobic and resistance training. Therefore, early treatment focused on the initial stages of sensorimotor training, including motor imagery. Pictures were taken of the patient, with the involved lower extremity in the gait phases in which the most significant deviation occurred (terminal swing and loading response). Based on the procedures used by Moseley,[24,25] the patient was instructed to imagine moving his limb into these positions during gait 10 times every waking hour. It was also recommended that he consider solutions to minimize the dysfunctional movement pattern to include a return to the use of crutches. Due to limitations of the patient's schedule and the recommended time frame for progression of motor imagery,[24,25] the next follow-up occurred 2 weeks later.

At the follow-up 2 weeks later, the patient indicated that his work schedule made it very difficult to adhere to the motor imagery and that it would not be practical to go back to crutches. The patient indicated that he would be more adherent to a program involving more movement-related tasks. Therefore, the patient skipped the initial recommended phases of imagery-based training[6,24,25] and was progressed to additional sensorimotor training activities, including graded movements with feedback based on knowledge of performance during non–gait-specific tasks. Based on impairments identified in the examination, the patient was instructed in the performance of great-toe tapping and rapid heel-to-toe tapping, with the goal of gradual progression of the performance of these tasks in a manner similar to that of the uninvolved extremity. He was also encouraged to obtain the brain-training program used in the Byl et al[6] investigation or similar commercially available software to fulfill the brain-fitness facet of the rehabilitative program. The patient was recommended to follow up in 2 weeks; however, work and family obligations prevented his adherence to the program and further follow-up.

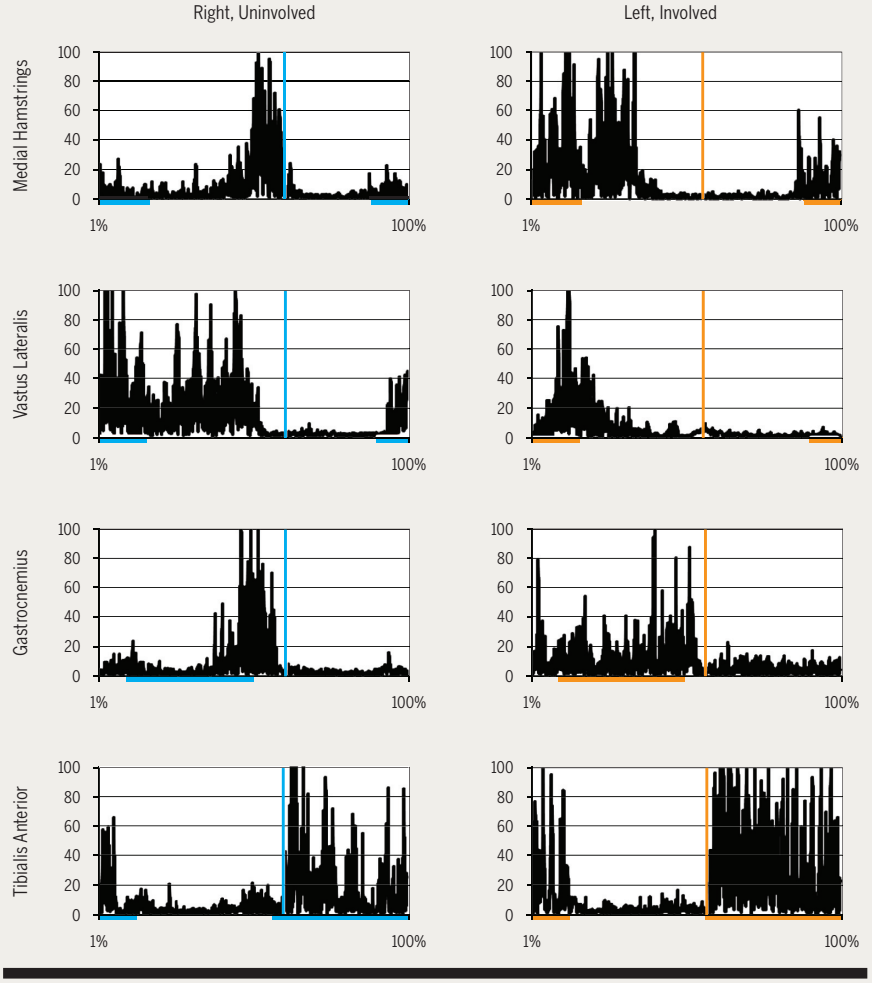

FIGURE 2. Surface electromyographic analysis revealed the following for the involved (left) side: (1) early and increased activation of the medial hamstrings during terminal swing and stance phases, (2) delayed and reduced activation of the vastus lateralis during terminal swing and stance phases, (3) cocontraction of the medial hamstrings and vastus lateralis during initial stance phase, (4) increased activation of the tibialis anterior during swing phase, and (5) early activation of the gastrocnemius during stance phase. The gait cycle is represented by initial foot contact (1%) and the point just before initial foot contact of the subsequent stride (100%). The vertical orange and blue lines depict toe-off for the left and right sides, respectively. Horizontal bars represent normal muscle-timing patterns during walking.[32] The vertical (*y*) axis represents full wave-rectified EMG signal normalized to the maximum activation of that muscle during the gait cycle. Abbreviation: EMG, electromyography.

DISCUSSION

Due to the reported low prevalence of runner's dystonia and any other form of primary task-specific lower extremity dystonia, misdiagnosis as an orthopaedic condition is common.[12,41] Task-specific dystonias have been associated with repetitive tasks, including sport- and instrument-specific activities that are prone to overuse and overpractice.[1] Increasing trends in running participation and high-mileage events may result in the right circumstances for increased observation of runner's dystonia. Further investigation is needed to accurately define the current prevalence of runner's dystonia.

Clinical Examination Considerations

The diagnosis of primary task-specific dystonia is currently based on an idiopathic onset of movement dysfunction specific to a task in the absence of other

TABLE 3 Sensorimotor Tests and Outcomes of Patient Evaluation Postdiagnosis of Runner's Dystonia Affecting the Left Side

Test	Description	Test Outcome	
		Right	Left
Digital reaction-time test[6]	Number of repetitions fully depressing a spring-loaded lever with the hallux in 60 s	191	159
Stereognosis[17]	Ability of patient to identify 13 common items with the foot presented in random order. Percentage of items identified incorrectly or partially incorrectly is reported (ie, percent error)	0% incorrect, 38.5% partially incorrect	7.7% incorrect, 46.2% partially incorrect
Kinesthesia[30]	Accuracy of response to movement of hallux in extension or flexion. Percentage of inaccurate responses is reported (ie, percent error)	0%	0%
Position sense[30]	Accuracy of response to position of hallux in extension, flexion, or neutral. Percentage of inaccurate responses is reported (ie, percent error)	0%	0%
Vibration[30]	Ability to detect vibratory versus nonvibratory sensation with tuning fork applied to the tibial tubercle and lateral malleolus with ears plugged. Percentage of inaccurate responses is reported (ie, percent error)	0%	0%
Semmes-Weinstein 10-g monofilament test	Ability of patient to detect buckling of a 10-g monofilament applied for 1 to 2 s to the plantar surfaces of the hallux, third metatarsal, and fifth metatarsal.[14] Additional testing of the plantar surface of the second and fourth metatarsal was performed. Percentage of inaccurate responses is reported (ie, percent error)	0%	0%
2-point discrimination	Smallest distance (in mm) to detect 2 stimuli applied to the tip of the hallux using a sliding 2-point discrimination tool with 1-mm increments. Normal, 6.6 mm[30]	6 mm	7 mm

neurological or orthopaedic disorders that would explain the presentation.[5,10,40] It has been suggested that runner's dystonia is a form of task-specific dystonia, with onset during running making this form distinct from other task-specific dystonias.[41] Currently, the diagnosis of task-specific dystonia is based on exclusion of competing diagnoses, yet technological advancements show promise in making a more direct diagnosis of dystonia.[38] In the case presented here, inconsistent orthopaedic and basic neurological examination findings relative to the gait dysfunction led to suspicion of dystonia as the cause of the patient's gait impairment. When compared to literature reports on runner's dystonia, the following features of the physical therapy examination were consistent with runner's dystonia: (1) task specificity of the movement dysfunction, (2) long history of running with first onset of symptoms during running, (3) inconsistent orthopaedic examination (muscle performance, coordination, joint mobility, and fractionation testing not consistent with the observed gait pattern), (4) negative basic neurological examination, (5) absence of pain, and (6) sustained and excessive muscular activation consistent with the altered gait pattern.

Additional features that can aid the physical therapist in diagnosing runner's dystonia include demonstration of sensory or motor tricks. Sensory tricks involve the use of touch to ameliorate the dysfunctional movement (eg, pressing on the hip while walking) and motor tricks involve voluntary movements (eg, walking backward).[5] Although this case did not clearly demonstrate a dramatic sensory or motor trick, the patient did mention improvement in gait while walking at the beach and placing pressure on the hip. Sensory and motor tricks are unique features of task-specific dystonia that can aid in the diagnosis of the condition but also can be useful as part of treatment. Interoceptive sensory tricks have been demonstrated to temporarily ameliorate dystonic movements of individuals with task-specific lower extremity dystonia, including 1 case of runner's dystonia.[20,36] If dystonia is suspected, the therapist should inquire about, and test for, sensory and motor tricks.

Sensorimotor examination of the patient with task-specific dystonia may reveal altered sensorimotor function, as demonstrated through testing of graphesthesia, stereognosis, kinesthesia, touch localization, motor accuracy/regulation, and spatial discrimination threshold.[2,10,22,23,29,33] In this case, the patient demonstrated impairments in fine motor control and stereognosis of the affected lower extremity. Adapted measures from sensorimotor testing in task-specific dystonia of the upper extremity used to test the lower extremity have not been defined in the literature. Further research is needed to assess the adaptability of sensorimotor tests used in the upper extremity to the lower extremity, in addition to the development of tests that can accurately detect lower extremity sensorimotor impairments.

Another consideration of sensorimotor testing in task-specific dystonia is that sensorimotor impairments may differ between the involved and uninvolved limb[22] but also may be demonstrated in both extremities.[10,23] In this case, discrepancies in sensorimotor impairments between the involved and uninvolved extremities

were the focus of the sensorimotor learning intervention. Comparisons of sensorimotor performance to the uninvolved extremity may not always be valid, considering that bilateral impairments have been demonstrated in unilateral upper extremity task-specific dystonia.[10,23] Additional research is needed specific to the lower extremity to improve confidence in results of sensorimotor testing used for diagnostic and treatment decisions in lower extremity task-specific dystonia.

Other factors that can be associated with task-specific dystonia include psychological and traumatic triggers.[1,5,18] By definition, primary dystonia includes dystonia without a known cause, whereas secondary dystonia includes physical or neurological trauma, medicinal or environmental toxicity, or psychogenic factors associated with the onset of dystonia. In the case presented here, a clear link to trauma or environmental or psychogenic factors cannot be made, but may have some association with the patient's symptoms and should be considered in the examination of a patient suspected of having dystonia. Although psychosocial inventories were not performed, it was evident from discussions with the patient's family that there was stress related to his work, coupled with a busy family schedule. Physical trauma or psychogenic factors may trigger dystonia or simply be a chance co-occurrence.[18] Psychogenic characteristics that have been associated with dystonia include perfectionism, social phobias, and other specific phobias (acrophobia, claustrophobia, etc).[1] Psychological or potential traumatic findings should be considered in conjunction with the physical therapy examination.

When clinical examination results suggest a task-specific dystonia, referral to a physician specialist in movement-related disorders is needed to confirm the diagnosis through additional testing. Multidisciplinary efforts are important in making the correct diagnosis and providing appropriate treatment recommendations.

Neurophysiological Testing

Further considerations beyond the physical therapy clinical examination to help in making the diagnosis of primary task-specific dystonia include brain magnetic resonance imaging to rule out structural lesions or metabolic disorders, genetic testing to identify presence of the DYT1 gene associated with early-onset generalized dystonia, and an ineffective therapeutic trial of levodopa to rule out dopa-responsive dystonia.[40] Currently, tests with known diagnostic standards that can provide further definitive information in altering the diagnostic probability of dystonia are lacking.

Although primary dystonia is idiopathic, there is evidence of abnormal neurological manifestations that may provide insight into the etiology of this disorder and the potential for development of diagnostic tests and treatments for dystonia. Tests such as transcranial magnetic stimulation, temporal discrimination threshold, and magnetoencephalogram reveal abnormal cortical representations in dystonia.[10,38] Paired associative stimulation and muscle vibration techniques reveal abnormal neural plasticity and reduced intracortical inhibition in dystonia cases.[37,38] Using transcranial magnetic stimulation, disruption of the sensory cortex organization and dystonic motor symptoms has been observed when primates are trained in a repetitive motor task for 12 to 25 weeks.[7,8] The sensory cortex disruption appears to result in convergence of adjacent areas of the cortical map that control the involved limb. Conversely, divergence of the cortical map through training has been observed to occur with the reduction of dystonic symptoms.[34] Although neurophysiological tests have demonstrated abnormal neurological findings in dystonia, clinical guidelines have yet to be established to apply these tests in its diagnosis. Given the complexity of the clinical presentation, the use of a cluster of tests and symptoms comprising items from the history or examination (eg, onset with repetitive tasks, absence of pain, sustained or excessive muscular activation, task-specific motor control,[10] and sensorimotor examination findings) and paired associative stimulation, transcranial sonography, or magnetoencephalogram results may better detect primary task-specific dystonia, yet further work is needed to assess the sensitivity and specificity of such testing.[38] In addition to diagnostic utility, further investigation of paired associative stimulation, transcranial sonography, or magnetoencephalogram may be valuable to correlate outcomes of interventions designed to modify cortical plasticity observed in task-specific dystonia.[10]

Treatment of Runner's Dystonia

The challenges associated with the diagnosis of dystonia can have significant implications on the dystonic patient's treatment plan and outcomes. The recommended treatment plan for dystonia is significantly different from the plan for common orthopaedic conditions for which it may be misdiagnosed. In this particular case, the patient's function declined significantly, as indicated by the decrease in LEFS score from 51/80 to 40/80 over the 10-month period that dystonia was not considered as a diagnosis. During this time, the patient received treatment including physical therapy and surgical intervention directed at orthopaedic conditions. Once the diagnosis of runner's dystonia was made, the treatment recommendations, based on a more accurate diagnosis, resulted in improvement of the patient's condition, reflected by an increase in the LEFS score to 55/80. This level of improvement exceeded the 9-point scale change indicative of the minimal clinically important difference of this measure.[4]

Cases of runner's dystonia reported in the literature have been managed with medication used in Parkinson's disease (levodopa, trihexyphenidyl), anticonvulsant medication (carbamazepine), neurotoxin injection (Botox), bracing, and mental imagery based on a sensory trick.[19,36,41] Each case demonstrated some

improvement but continued to demonstrate impairment and significant limitation of running, despite treatment.

Currently, the most common treatment for any type of lower extremity dystonia includes Botox and trihexyphenidyl.[21] Only 2 cases were found describing nonmedicinal or noninvasive management of task-specific lower extremity dystonia.[3,36] In 1 case, functional electrical stimulation was shown to improve but not fully resolve the patient's gait dysfunction.[3] In another case of an individual diagnosed with runner's dystonia, treatment included the use of mental imagery based on a sensory trick and reduced the dystonic symptoms.[36] Longer-term treatment and outcome information of this latter case was not reported, yet imagery based on a sensory trick may be an effective short-term treatment as a component of the patient's overall, longer-term management.

Due to the lack of evidence for task-specific lower extremity dystonia, the initial treatment plan in this case was based on literature from task-specific upper extremity dystonia. Sensorimotor testing and interventions used for upper extremity dystonia may not directly translate to the lower extremity, due to the different motor competencies between the hand and the foot. In addition, the validity and reliability of the adapted sensorimotor tests used in this case may not reflect test properties as originally demonstrated and therefore may not be as effective in decision making when applied to the lower extremity. Finally, current sensorimotor-based treatment programs are very time intensive and in this case proved to be prohibitive to the patient's ability to complete treatment.

Task-specific lower extremity dystonias affecting gait pose additional problems with sensorimotor training compliance, particularly the avoidance of the offending movement. Avoiding or altering gait to reduce the dystonic movement affects the patient's ability to maintain social roles in a manner different from upper extremity dystonias (eg, writer's cramp, musician's dystonia). The ability to achieve improvement or resolution of task-specific lower extremity dystonia with less time-intensive training or without cessation of the dysfunctional pattern has not been demonstrated, yet current recommendations include aversion of the offending movement pattern and frequent sensorimotor-based training.[6,24,25] In addition, the effect of bypassing initial imagery phases for the sake of compliance is not known, although interpretation of current evidence using sensorimotor training can only be applied within the context of the recommended progression, including imagery training. Functional electrical stimulation may be another option in conjunction with sensorimotor and medical management of dystonia, yet only 1 case has been presented using this treatment for lower extremity dystonia.[3] Further research is needed to define the most effective strategies for physical therapy management of lower extremity task-specific dystonia.

CONCLUSION

Runner's dystonia is a rare yet possibly underdiagnosed condition that results in abnormal movement patterns inconsistent with physical and basic neurological examination results. In the case presented here, dystonia was not initially considered in the differential diagnosis, despite evaluations by multiple healthcare specialists. Once the diagnosis of runner's dystonia was made, the treatment recommendations shifted toward interventions directed at the altered supraspinal control of the motor system, sensory perception/integration, and sensorimotor integration.

Current evidence indicates the following features that can be assessed by the physical therapist in support of the diagnosis of runner's dystonia: (1) task specificity of the movement dysfunction, (2) long history of running with first onset of symptoms during running, (3) inconsistent orthopaedic examination, (4) negative basic neurological examination, (5) absent or minimal pain, (6) sustained or excessive muscular activation consistent with the altered gait pattern, (7) sensory or motor tricks, and (8) sensorimotor impairments. Diagnosis and management should include a multidisciplinary effort to include physical therapy and a physician specialist in movement-related disorders. Physical therapists can provide sensorimotor training in addition to physical and brain-fitness training that can be facilitated by medicinal and/or injection-based interventions to reduce the dysfunctional movement pattern. Further research is needed to demonstrate effectiveness of treatment for runner's dystonia. ◉

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