ORIGINAL ARTICLE

Ballism After Stroke Responds to Standard Physical Therapeutic Interventions

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ABSTRACT. Mark VW, Oberheu AM, Henderson C, Woods AJ. Ballism after stroke responds to standard physical therapeutic interventions. Arch Phys Med Rehabil 2005;86: 1226-33.

Objective: To report the effects of noninvasive standard physical therapy (PT) interventions on an involuntary movement disorder after stroke.

Design: Single case with clinical follow-up over 2 years.

Setting: Inpatient stroke and rehabilitation services and outpatient clinic.

Participant: A man with acute bilateral ballism after unilateral subthalamic infarction.

Interventions: Rhythmic coordinated bilateral limb movements and firm tactile stimulation to the hand.

Main Outcome Measures: We had not anticipated that dyskinesia itself would specifically improve during treatment. Consequently, we used qualitative clinical observations, including review of videotaped performance, and self-reported limb control. Hypotheses concerning treatment effects were developed after data collection.

Results: Involuntary movements recurrently improved within treatment sessions.

Conclusions: PT may improve dyskinesia after stroke. The benefit may be adjunctive or alternative to current invasive treatments of movement disorders after brain injury and merits confirmation. The improvements are consistent with current research indicating that (1) intact cortical, subcortical, cerebellar, and spinal areas interact to generate bilateral rhythmic limb movements that can overcome dyskinesia and (2) tactile stimulation can improve motor deficits associated with basal ganglia disorders. Finally, because functional activities assessments improved our evaluation of ballism, these should be routinely used along with conventional neurologic examination to assess involuntary movement disorders.

Key Words: Activities of daily living; Ballismus; Cerebrovascular accident; Rehabilitation; Subthalamic nucleus.

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HEMIBALLISM IS AMONG the most impressive of the rarely encountered "hyperkinetic" movement disorders that can follow cerebral infarction,¹ consisting of the involuntary, erratic flinging of the proximal limbs on 1 side. It is most commonly associated with a lesion in or near the contralateral subthalamic nucleus,² although other solitary lesion sites have also been implicated.³ The bilateral presentation of this movement disorder, which may be simply termed ballism, is less often described.^{4,5} Clinical reports^{3,4,6,7} of ballism with neuroanatomic findings have generally indicated bilateral focal structural lesions or diffuse injury. However, a few case reports^{4,8} have associated ballism with strictly a unilateral lesion.

Case studies seldom describe the functional effects of ballism. Self-inflicted soft-tissue or skeletal injuries have been reported.^{4,7,9-12} Some studies^{9,13} have indicated interference with eating, writing, speaking, buttoning, and walking, but functional assessment tools were not indicated. Published treatments of persisting ballism have been limited to pharmacologic or surgical interventions^{14,15} and have not evaluated responses to physical therapy (PT). Although specific medications are often effective, side effects frequently occur.14 Stereotactic neurosurgery (eg, pallidotomy) is reserved for refractory cases and is highly successful for ameliorating symptoms yet is associated with residual functional impairment in 30% of patients.¹⁵ Ballism of vascular etiology resolves within several months in most cases, with or without invasive treatment.¹ However, patients with ballism after stroke have an increased risk of stroke recurrence and mortality, as is consistent with stroke in general. One study¹⁶ found a survival rate after vascular ballism of 51% at 36 months and 32% at 150 months.

We recently observed acute bilateral ballism after unilateral subthalamic nucleus infarction in a patient. We followed the movement disorder's interference with his functional activities over 2 years and report the favorable response to noninvasive therapeutic interventions, which we had not anticipated. Because we did not have access to physiologic instruments that could have objectively assisted this evaluation (eg, movement analysis kinematics), our report relies on clinical observation and self-report. Nonetheless, our observations suggest that noninvasive rehabilitation techniques may at least temporarily ameliorate the symptoms, which could improve treatment participation and outcomes. These findings merit verification in other patients with ballism.

METHODS

Acute Patient Presentation

A 66-year-old right-handed retired salesman, previously functionally independent, awoke one morning with right-sided involuntary limb jerks that were precipitated by intentional movement, affecting the arm more than the leg. He also noted impaired speech fluency and word retrieval. He was admitted to our institution's acute stroke service on the same day.

The patient's medical history included multiple vascular disease risk factors (diabetes, hypertension, hyperlipidemia, recurrent myocardial infarction, smoking) and several episodes of left facial weakness during the preceding 20 years that had

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Fig 1. MRI sequences from the patient. (A–C) Consecutive FLAIR sequences. Arrow indicates abnormal signal. (D) FLAIR sequence above the lateral ventricles. (E) Diffusion-weighted sequence at the same level as B. (F) Adjusted diffusion coefficient map at the same level. Division marks equal 1cm.

lasted up to a few days each. The most recent cerebral magnetic resonance imaging (MRI) study from 2 years before the present admission (when the patient had last experienced facial weakness) had shown microangiopathic changes but no infarction.

The patient's admission medical examination indicated bilateral involuntary limb movements (detailed below), modest right hemiparesis (diffusely 4/5 power by Medical Research Council criteria¹⁷), and a 50% reduction in light touch appreciation on the right hand and face when compared with the left side. Joint position sense and pin appreciation were preserved in all limbs. No other cranial nerve deficits were present. Confrontational visual fields to finger counting were full without extinction. Myotatic reflexes¹⁸ were brisk on the right, normal on the left. Plantar responses were flexor. Admission blood test results were unremarkable apart from moderately elevated serum glucose (182mg/dL). The chest radiograph examination result was normal.

MRI on the day of admission showed a bright signal in the left subthalamic nucleus on the fluid-attenuated inversion recovery (FLAIR) sequence (figs 1A–C), as well as extensive bilateral supraventricular white matter disease (fig 1D). The diffusion-weighted imaging (DWI) sequence and apparent diffusion coefficient maps indicated signal changes only in the left subthalamic nucleus (figs 1E, 1F). These findings were compatible with acute ischemia.¹⁹

Transesophageal echocardiography showed a dilated left atrium (43mm); thickened, nonstenotic mitral and aortic valves; and normal ejection fractions. Four-vessel angiography indicated a normal right carotid system, with persistent fetal origin of the right posterior cerebral artery and mild stenosis at the origin of the left internal carotid. The transcranial Doppler examination result was completely normal, with no evidence of hemodynamic abnormalities.

Rehabilitation Course

The patient's neurologists decided against further intervention other than to add aspirin and clopidogrel to his medications. The patient was then transferred to our acute inpatient stroke rehabilitation service on day 6 of his illness. His impairments included mild word-finding deficits, impaired short-term verbal memory, mild dysarthria, profound ambulatory deficits, and inability to perform self-care tasks safely. The patient's wife thought the cognitive impairments originated with this illness and have continued to the present. Formal neuropsychologic assessment yielded a normal score of 27 out of 30 on the Mini-Mental State Examination,²⁰ with deficits restricted to delayed verbal recall. On the Boston Naming Test,²¹ the patient named 54 out of 60 items, normal for his age.²² Tests of limb praxis that were conducted after hospital discharge, while he was still symptomatic from his movement disorder, showed normal performance on the Florida Apraxia Screening Test pantomimes to command,²³ the imitating of meaningless hand postures,²⁴ and the reproduction of meaningless limb movement sequences.²⁵ At no time were complex involuntary movements observed, such as mirror movements, disinhibited grasping, compulsive tool use, or intermanual conflict.

The patient consented to videotaping and investigational evaluation of his motor progress during inpatient and outpatient rehabilitation, as approved by our institution's human subjects research board. Involuntary movements seldom occurred at rest, but attempts to actively move the right arm invariably caused wide, erratic, and rapid gyrations of the proximal limb, such that he once struck his face and repeatedly bruised his right arm. The nondominant left arm displayed no such movements while the patient was supine at rest. However, simulated shaving by his left hand with an unplugged shaver produced jerky but noninjurious movements about the face. Rapid alternating movements (supination and pronation) and finger-tonose touching were bilaterally clumsy, right much more so than left. The left hand also showed past-pointing that resolved with a few reattempts. Occasional isolated jerks of the trunk appeared during limb movements. Although stuttering and wordfinding delays disrupted speech, there were no involuntary movements of the eyes, face, oropharynx, or neck.

The bilateral reciprocal forearm rolling test²⁶ was used to evaluate motor control. The test simply requires rapid, antiphase cycling of both forearms in the same direction in the midsagittal plane about a common focal point without synchronization to an external stimulus. Initially he showed wild, dangerous excursions by the right arm and smaller movement amplitude with the left. However, after continuing the maneuver for half a minute, the arm movements unexpectedly equalized, so that the right arm moved with reduced amplitude and jerkiness, while the left arm amplitude increased. The movement disorder immediately worsened with reversed arm rotation, but again subsided with continuation.

Initially, the patient required moderate to maximal assistance to come from sitting to standing and maximal assistance to use a hemi-walker, because of interference from the concurrent irregular movements. When he walked with 2 therapists as they gripped his hands for support, all limbs showed proximal ballistic movements at various times, more so on the right. When he walked with a single-point cane in the right hand, bilateral contact guard assistance of 2 therapists was required, because the right hand and leg and his trunk shook severely. Unexpectedly, however, when he supported himself with the cane in his *left* hand, he required contact guard assistance from only 1 individual, because the ballism had diminished. These findings were reproducible.

The patient's ambulatory training involved wearing a 5-lb (2.3-kg) weight on his right leg and manually restraining the right upper limb as he walked with a rolling walker. Nonetheless, flailing movements occurred repeatedly, requiring maximal assistance to maintain his safety. By day 8 after stroke



Fig 2. Signatures from the patient according to day of illness and hand. Only the top halves of the signatures are shown to conceal the patient's identity. All samples are to the same scale. Measurement bar equals 5cm.

onset, the movement disorder had reduced in weight-bearing positions, such as when grasping a hemi-walker. Having the patient walk in an intentionally rhythmic manner markedly reduced the hyperkinesia within the therapy sessions. The ballism gradually subsided during the hospital course.

Self-care activities were minimally impaired, but the patient consistently relied on his nondominant left upper limb. Reaching was practiced with a 5-lb weight on the right wrist; however, the right upper limb would flail when he used an open-chain movement sequence (absence of fixed distal limb contact with a surface for weight bearing).²⁷

Serial evaluation of our patient's handwriting with both hands (fig 2) showed that early in his illness he was completely unable to put pen to paper with his right hand. His left hand could sign his name legibly but with an irregular speed and large strokes. The irregular motion of the right hand during signing could not be ameliorated by having his left hand simultaneously grip an object (a different pen). We did not assess signing with the right hand while the left hand was engaged in a closed-chain activity. During subsequent days both hands improved in control and amplitude. The patient's initial FIM instrument²⁸ score was 57, and by

The patient's initial FIM instrument²⁸ score was 57, and by discharge on the eighth inpatient rehabilitation day it had improved to 82 (maximum score, 126). By this time he still primarily used his nondominant hand for self-care, because fine motor function in the right hand remained impaired. He required only minimal assistance from his wife to walk with a standard rolling walker or to climb stairs, but he had to be cautious, because ambulation without assistance provoked right-sided ballism, especially in the lower limb. Similar to earlier observations, the ballism abated when he gripped the examiner's hand with his left hand or gripped a walker with both hands but not when he gripped the examiner's hand with his right hand. Coming from sitting to standing without assistance provoked truncal jerking, which required minimal to moderate assistance for stability, but once up, he could safely commence walking.

We treated the patient with 50mg/d of sertraline because of its reported rapid and well-tolerated benefit for hemiballism.²⁹ However, the severity of the movement disorder failed to improve over a few months after hospital discharge while on this medication.

Outpatient Course

The patient had outpatient therapy 3 hours a week for 1 month. Initially, self-care activities were performed primarily

with the left hand. He was noted to have a pronounced startle affecting the trunk and proximal limb muscles when he was surprised by visual or auditory stimuli. By this time he could walk unassisted without tremor and did not need an assistive device. The forearm rolling test was performed flawlessly. Writing had improved such that his right-hand signature was better controlled than that of the left hand (see fig 2). Although he could feed himself independently, irregular proximal arm tremors persisted on tasks that involved repetitious fine distal movement of the right hand, particularly when he felt fatigued. The facial and right-hand sensory impairments did not improve. By the end of outpatient therapy he had resumed his pastime of fishing, and in the clinic he competently demonstrated casting with a rod and reel without losing standing balance. He also resumed driving without difficulty. The patient was satisfied with his progress and terminated therapy.

The movement disorder and hemisensory deficit were acutely exacerbated 5 months after illness onset, but he declined hospitalization. Conventional MRI studies at 6 and 10 months were unchanged. In our outpatient clinic at 6 months, his speech was more dysfluent, and ballism was present in his right upper extremity at rest. Because the patient had concurrent depression that had not responded to sertraline, the sertraline was changed to 75mg/d of venlafaxine (which, like sertraline, inhibits serotonin reuptake, but additionally inhibits norepinephrine reuptake). The depression subsequently improved, but the movement disorder did not. On the 30-item Motor Activity Log (MAL)³⁰ amount of use scale, he rated his right arm function at a mean of 3.6 and 3.1 (maximum score, 5.0) at 7 and 8 months into his illness, respectively, which indicates moderate nonuse. At 10 months after illness onset his stroke neurologist started him on 1mg of haloperidol twice daily. Consistent with earlier reports,^{16,31} ballism severity was reduced but not abolished. His MAL score improved to 4.3. At 11 months, a neurologist substituted 1mg of clonazepam 3 times a day for the haloperidol to avoid tardive dyskinesia, and the patient noted further reduction of symptom severity. When evaluated at 17 months, he indicated that he could control right-sided involuntary movements by clenching a pocketknife in his right pocket, and rhythmic walking continued to benefit his gait. Postural stability during walking was better controlled with a single-point cane in his left hand than in his right. Speech remained hesitant. The patient terminated physiatric follow-up at this time because he felt his recovery had plateaued.

On informal follow-up at 24 months after illness onset, he indicated that the movement disorder was overall unchanged but often fluctuated, in particular becoming aggravated when he was in crowded settings such as church. He had experienced 3 falls from sudden dyskinesia-associated imbalance in the preceding 3 months, fracturing his right shoulder at one point. He reported no further improvement from the clonazepam and continued to ameliorate his involuntary movements by grasping a pocketknife with his right hand. He continued to be followed up by his stroke neurologist twice yearly, but without further medication changes.

DISCUSSION

Our case has several notable findings. One, detailed neuroimaging suggested an acute unilateral infarction that apparently precipitated bilateral ballism. Two, the functional impact of the ballism is described in considerably more detail than in previous reports. Three, the ballism was ameliorated by noninvasive therapeutic techniques, which has not been reported.

Neuroimaging Findings and Anatomic Relationships

DWI is more sensitive than conventional MRI to cerebral infarction.³² Our patient apparently had acute infarction confined to the left subthalamic nucleus, despite bilateral motor signs. Previous studies of bilateral ballism did not use this imaging technique, although patients with unilateral ballism that follows marked hyperglycemia have shown DWI findings similar to ours.³³ The symptom exacerbation that occurred at the fifth month of the illness was most likely caused by recurrent subthalamic infarction that was not visible on standard MRI.

Most contemporary reviews of the subthalamic nucleus's contribution to purposive movement indicate only its ipsilateral connections with other basal ganglia.³⁴⁻³⁷ Hemiballism that follows subthalamic nucleus lesions is thought to result from loss of excitatory drive to the ipsilateral medial globus pallidus. This, in turn, disinhibits the ipsilateral ventrolateral thalamus and thus unleashes excessive cortical motor activation of the contralateral limbs. However, this model does not explain how a unilateral cerebral infarction would precipitate a bilateral hyperkinetic disorder.

In contrast, other investigators have indicated that the subthalamic nuclei are also interconnected across the body midline via an interhemispheric commissure.^{6,8,38,39} The excitatory subthalamic nucleus efferents are glutamatergic.⁴⁰ Thus, injury to 1 nucleus could reduce excitation to the opposite nucleus, resulting in bilateral but asymmetric ballism, as shown by our patient. In the marmoset, unilateral subthalamic ablation results in bilateral neurochemical changes in the basal ganglia that are thought to be mediated by commissural relays.⁴¹ Unilateral subthalamic stimulation to relieve Parkinsonism primarily has contralateral benefits, but nonetheless mild improvement of ipsilateral motor deficits also results.⁴² It has been suggested that the bilateral supplementary motor area activation that occurs with unilateral subthalamic stimulation for Parkinsonism⁴³ reflects relays between either the globus pallidus internus or the substantia nigra and the contralateral thalamus.44 Regardless of the specific pathways involved with bilateral ballism, these studies support our impression that acute unilateral subthalamic injury in humans can have bilateral motor effects. It is also conceivable that the extensive bilateral white matter ischemic changes suggested by our patient's MRI scan may have contributed to the bilateral presentation of ballism after unilateral infarction.

In contrast to many other cases of ballism,³ the disorder was rarely present at rest and did not involve the face. Consequently, our patient manifested relatively mild ballism, the self-injuries notwithstanding. At times the upper limbs (particularly the left) also presented classic cerebellar disturbances (past-pointing, clumsy alternating movements). These disturbances may have resulted in part from the infarction's involving the dentatorubrothalamic tract,⁴⁵ which courses close to the subthalamic nucleus.⁴⁶ Frequently, ballism is not "pure" but is instead associated with other involuntary movements.^{3,47} Thus, the co-occurrence of diverse movement disorders in our patient is consistent with other reports of ballism.

Results of Functional Assessments

Unlike previous reports of ballism, we extensively assessed functional activities. Bilateral ballism was more evident during functional activities (walking and writing in particular) than on standard bedside neurologic evaluation. Consequently, the omission of functional activities assessments from movement disorder studies may preclude identifying bilateral ballism. The additional sensitivity of functional assessment beyond laboratory or simple clinical evaluations has been noted for unilateral spatial neglect^{48,49} and Parkinson's disease.⁵⁰ Therefore, the infrequency of reports of bilateral ballism, relative to hemiballism, may be because conventional neurologic examination is insensitive to its bilateral but asymmetric presentation. Similarly, bilateral motor deficits are well known to follow unilateral cerebral infarction associated with clinical contralateral hemiparesis, but special testing or functional evaluation are usually required to uncover the ipsilesional deficits.⁵¹

Our patient showed moderate nonuse of the more-affected upper extremity—that is, reduced spontaneous use of the limb on functional tasks, despite intact movement to command—based on a validated self-report instrument (ie, the MAL).³⁰ Previous reports of nonuse after stroke have assessed hemiparesis but did not indicate limb hyperkinesia. Our report thus shows that moderate nonuse of the more-affected upper extremity after stroke may include asymmetric hyperkinesia. As has been hypothesized for other cases of nonuse after neurologic injury, the nonuse of our patient's more-affected arm was probably learned,⁵² secondary to adverse conditioning from attempted movement.

Responses to Noninvasive Interventions

As was seen in our patient, vascular ballism commonly subsides within weeks of cerebral infarction. The various medications that were used to treat his movement disorder at best only partially ameliorated the symptoms. We cannot exclude the possibility that higher doses might have been more beneficial. However, after our initial medication attempt to treat the movement disorder, we favored treating the patient's depression because (1) we were concerned about possible side effects from increasing doses of existing medication, (2) the patient had behaviorally adapted to his movement disorder (as has been reported for ballism¹⁵), and (3) we did not want to neglect his depression. The depression responded quickly and impressively to venlafaxine,⁵³ which was thereafter chronically continued without discernible effect on the ballism.

Because our patient's ballism incompletely responded to medication, it is important to consider whether the favorable results that apparently followed the standard stroke rehabilitation techniques we used suggest that these approaches may benefit ballism in general. These interventions—continuous repetitive movement (rhythmic walking, forearm rolling) and partial weight bearing (closed-chain activity) or gripping an object with either hand—generally improved control of the movement disorder except when the patient was fatigued. As a result, locomotion stabilized and could be more easily treated. These improvements may have facilitated PT for functional motor recovery.

The physiologic bases for these improvements are unclear. However, several lines of evidence from related disorders and studies in healthy subjects suggest contributing mechanisms.

Rhythmic, synchronized bilateral limb movement. Patients with neurodegenerative movement disorders (Parkinson's disease, Huntington's disease) improve ambulation when they synchronize their walking to environmental rhythmic stimuli.^{54,55} Similarly, patients with diverse cerebellar disorders can overcome action tremor of the upper extremity with continuous, self-developed rhythmic limb movements.⁵⁶ Physiologic neuroimaging studies suggest that an extensive network of neural structures collaborates in the motor learning of repetitive limb movements, including the cerebellum, sensorimotor cortex, premotor and supplementary motor areas, and striatum (caudate and putamen).⁵⁷ Central pattern generators in the spinal cord are also postulated to mediate bilaterally coordinated rhythmic behavior.^{58,59} The premotor, dorsolateral pre-

frontal, and parietal areas may be particularly involved when coupling the limbs during self-developed repetitive movements, unlike when synchronizing the limbs to rhythmic environmental stimuli.⁶⁰ This suggests that attentional and executive mechanisms are recruited for self-paced rhythmic movements. As learning a self-paced, reciprocal bimanual task proceeds, the activation of dorsolateral prefrontal and parietal areas diminishes,⁶¹ which suggests reduced attentive monitoring during procedural learning.

Within-session improvement when learning tasks,62,63 as was shown by our patient, is consistent with a 2-phase model of skill acquisition: first, a rapid within-session improvement, followed by a slower, continued improvement with practice over several days.⁶⁴ Neuroimaging studies⁶⁴ in humans suggest that corticocerebellar loops are recruited early during motor learning, whereas corticostriate loops may be preferentially activated later as the individual becomes proficient at a particular skill. However, when humans must adapt to perturbations from external forces on the limbs, the opposite order of anatomic activations has been described.⁶⁵ Thus, the specific sequence of regional brain activation may be task dependent, in part affected by whether the person must adapt to instability in limb movements.⁶⁴ The dyskinetic movements of ballism are similar to external forces on the limbs, because they challenge the individual to rapidly adapt to uncontrollable and abrupt perturbations, as indeed our patient did.

In addition, the corpus callosum and the supplementary motor area appear necessary for interhemispheric inhibition as a person learns reciprocal bilateral limb movements, ⁶⁶⁻⁶⁸ as well as to facilitate bilateral coordination. ⁶⁹ Lesions to these areas result in synkineses (mirror movements) or intermanual conflict. These abnormal movements did not appear in our patient, which along with our apraxia assessments suggested functional integrity of these structures.

The contribution of the subthalamic nucleus to procedural learning is unknown, probably because (1) its small size might be difficult to resolve during functional imaging studies, (2) ballism is uncommonly reported, and (3) motor learning in ballism has not previously been addressed. Nonetheless, the foregoing studies suggest that patients with ballism that follows subthalamic nucleus lesions may improve bilateral rhythmic limb movements with practice, because of the functional integrity of cortical, striatal, and cerebellar areas. The white matter abnormalities seen in our patient were apparently not sufficient to prevent these improvements. Because we evaluated only a single patient, we cannot indicate whether the reduction in dyskinesia in the weeks after the initial stroke's onset had been directly due to motor training effects rather than spontaneous recovery.

Somatosensory stimulation. The movement disorder also improved with partial weight bearing by the upper extremities or gripping an object within a physically restricting space (a pocket). In contrast, initiating limb movements without weight bearing, grasping an object without constraining movement (eg, using a pen), or even simply imagining the right arm moving induced ballism. Hence, improved control apparently was related to substantial somatosensory stimulation, at least to the hands.

Converging evidence implicates the basal ganglia in gating somatosensory information, particularly proprioception, which may be fundamental to the pathophysiology of diverse involuntary movement disorders⁷⁰⁻⁷² as well as their responses to peripheral stimuli. Substantial cortical somatosensory projections reach the basal ganglia via the ipsilateral subthalamic nucleus and putamen.^{73,74} The basal ganglia are active primarily during closed-loop limb movements (ie, those that are modified by afferent feedback⁷⁵), whereas the cerebellum and motor cortex are active during both closed-loop as well as open-loop ballistic movements that rely less on feedback.^{76,77} Consequently, basal ganglial injury may preferentially impair proprioception and hence the monitoring of slow limb movements, whereas ballistic movements may be spared. Subtle proprioceptive disturbance has been found in Parkinson's disease,⁷⁸⁻⁸⁰ which is considered primarily a basal ganglia disorder. Compensation for Parkinsonian akinesia or bradykinesia by environmental (visual or auditory) cuing⁸¹ may recruit relatively spared sensory channels to improve motor control.82 Choreoathetosis has been shown to follow proprioceptive loss from lesion anywhere in the nervous system, which may reflect aberrant sensorimotor integration in the basal ganglia.⁸³ The subthalamic nucleus is especially responsive to proprioceptive input relative to other somatosensory (eg, cutaneous) stimuli.⁸⁴⁻⁸⁶ It is therefore not surprising that experimentally induced ballism in monkeys can be aggravated by dorsal rhizotomy, which abolishes peripheral somatosensory input.⁸⁷ In a case report of human hemiballism, microelectrode recording showed reduced activation of the globus pallidus by proprioceptive stimulation, relative to healthy monkeys and patients with other involuntary movement disorders.84

The foregoing suggests that our patient's acute right facial and upper-extremity somatosensory loss resulted from his contralateral subthalamic infarction. Our failure to find proprioceptive disturbance may have been due to the crude manner by which standard neurologic examination assesses joint position sense in the fingers and toes.

Dystonias are similar to ballism in that they involve disinhibited muscular contraction,⁸⁸ although the contraction in dystonia is prolonged, whereas in ballism it is brief. Dystonias often improve with a "sensory trick" (geste antagonistique), which usually involves firm contact between the affected body part and another object, such as a bed or the patient's own finger.^{38,89-92} Thus, dystonias resemble Parkinson's disease in their benefit from external stimulation. The basis for improvement after sensory tricks is not completely understood. However, several basal ganglia disorders (cervical dystonia, Parkinson's disease, Huntington's disease) show ipsilateral sensory cortex activation rather than the usual contralateral activation when patients are touched on the more-affected side of the body.^{70,91} This shift may reflect compensation for contralateral basal ganglial somatosensory dysfunction, such as by reduced transcallosal inhibition of the ipsilateral hemisphere. In cervical dystonia this shift may help to counteract torticollis.

The effect of external stimulation on ballism, in contrast, has not been systematically evaluated. One case study93 noted temporary amelioration of ballism after applying cutaneous electromyographic electrodes. Early in his illness our patient benefited from left-hand weight bearing, but right-hand weight bearing aggravated his symptoms. Later in his illness, when ballism was less severe, clutching an object with the right hand in his pocket improved motor control further. These observations suggest that increased tactile contact reduced involuntary movements, but only when such movements were not already severe in the limb that attempted tactile contact. Consequently, when the less-involved left hand adopted weight bearing early in the illness, the right hemisphere may have been preferentially activated, which in turn may have helped to suppress the left hemisphere's disinhibited motor cortical activity through transcallosal inhibition. Later in the illness, when ballism was less severe, right-hand tactile stimulation may have benefited the movement disorder, because partial recovery of the subthalamic nucleus may have allowed improved proprioceptive processing from the right hand and hence reduced contralateral

motor cortical excitation. Alternatively, as shown in cervical dystonia and Parkinson's disease, tactile stimulation to the more-affected side of the body may have preferentially activated the ipsilateral sensory cortex, which may have second-arily inhibited the contralateral motor cortex through transcallosal inhibition. This would not have been possible earlier in the illness because of the severity of the ballism in the right upper extremity.

Our patient had excessive startle to unexpected abrupt visual or auditory stimuli. This phenomenon has not been reported in ballism. Experimental ablation of the subthalamic nucleus in animal models induces impulsivity and hyperactive complex behaviors such as feeding and locomotion.^{41,94-97} These findings suggest that the subthalamic nucleus has a general inhibitory function. The basal ganglia are hypothesized to participate in a general frontothalamic loop of functionally segregated circuits that mediate motor, cognitive, and affective processes.⁹⁸ Disruption of the "limbic" loop would result in impaired response inhibition or general hyperexcitability,99 as was seen in our patient. Cognitive or affective interactions with our patient's movements were reflected in the aggravation of his ballism when he simply imagined purposive limb use or had to mingle with crowds. These activities may have stressed attentional resources that were important for suppressing unwanted movements. Many case reports of ballism have indicated its aggravation during emotional arousal or concurrent cognitive activities (eg, speaking),^{4,7,8,10,12,93,100,101} whereas, in contrast, considerable attentional effort or, paradoxically, relaxation can suppress the involuntary movements.9,102

CONCLUSIONS

Although hyperkinetic movement disorders after stroke are apparently much less common than hemiparesis, they may be similarly disabling. Unfortunately, rehabilitation approaches for movement disorders in stroke are less well developed than for hemiparesis. Furthermore, rehabilitation approaches to involuntary movement disorders in general have seen little progress^{50,103} compared with invasive pharmacologic and surgical treatments, despite some promising indications. Our patient with bilateral involuntary hyperkinesia responded favorably to standard stroke therapy, at least within treatment sessions. Our interventions were initiated before our literature review and thus were not informed by diverse research findings in motor physiology. The improvements that were seen are consistent with different lines of experimental and clinical evidence. We hope that our findings will encourage other rehabilitation programs that may treat patients with acquired involuntary movement disorders to evaluate the benefit of our approaches.

Our clinical, neuroimaging, and therapeutic observations invite new questions about such disorders. In particular, it is important to know whether ballism consistently responds to the noninvasive rehabilitation interventions we have identified and whether the severity of the disorder affects outcomes. Because of the infrequency of ballism, it would be impractical to conduct extensive controlled clinical trials. However, further detailed case studies that would systematically control noninvasive interventions would contribute invaluable expertise. A national stroke registry to identify involuntary motor disorders would help to acquaint clinicians with their existence and foster information exchange that could expedite developing specific treatments. These findings would help to determine whether medications or neurosurgery are essential to control such disorders.

In addition, ballism may be underappreciated during conventional neurologic examination. Anatomic-functional studies of ballism should include standard assessments of daily living activities. Such investigations would help to clarify the contributions of the subthalamic nucleus and related structures to the control of voluntary movement and cognition and their associated disorders.

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