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Letter to the editor

Tibialis anterior tendinopathy in a dystonic talipes calcaneovarus foot: A case treated by botulinum toxin

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Dear Editor

Traumatic brain injury (TBI) often results in neuromotor disorders, particularly muscle overactivity. Although many of the manifestations of muscle overactivity during gait depend on the cycle phase and are non-permanent, sustained dystonic movements or postures can result in mechanical overload of tendons. Along with repetitive microtrauma, this mechanical overload is considered a leading cause of chronic tendinopathy — a frequent concern in work-related musculoskeletal disorders or in athletes. In contrast, tendinopathy has not been well characterized in neurological patients presenting muscle overactivity, especially those with muscle dystonia. Likewise, the value of botulinum toxin (BoNT) for treating tendinopathies in a context of muscle overactivity remains uncertain. Here, we report on a patient with traumatic brain injury (TBI) presenting dystonic talipes calcaneovarus and treatment-refractory tibialis anterior (TA) tendinopathy that was relieved by intramuscular BoNT injections.

A 41-year-old woman had severe TBI (with bilateral damage to the frontal and cerebellar regions of the brain) sustained in a car accident at age 23. Right hemiparesis and moderate cognitive disorders were still present when she was referred to our department. After the initial post-TBI rehabilitation phase, the woman had been able to walk (using a cane) over a distance of more than 2 km. However, a right talipes calcaneovarus deformity due to sustained activation of the foot dorsiflexors (and particularly the TA) had developed in both the swing and stance phases of gait. The stance phase had also been perturbed by ankle clonus related to hypertonia of the triceps surae and tibialis posterior (TP). Thus, tibial nerve neurotomy (involving the branches innervating the medial and lateral heads of the gastrocnemius, soleus and TP) was performed in 2008. This operation relieved the clonus but of course not the talipes calcaneovarus deformity during gait.

The patient began to complain of mechanical midfoot pain 18 months before she was referred to our department. She experienced pain after walking for more than 30 min and therefore after walking any distance. The pain was felt primarily in the medial and dorsal parts of the midfoot and radiated to the anterior part of the ankle. Paracetamol, tramadol and local physiotherapy (cold therapy, infrared therapy, and deep transverse friction massage) failed to provide sufficient pain relief. She received no local procedures (e.g., corticosteroid injections). The pain limited the woman's walking distance to 100 m and restricted several activities of daily living.

The patient's gait was characterized by impaired knee flexion in the swing phase and a sustained talipes calcaneovarus deformity. The lateral part of the right heel struck the ground first, and there was no first plantar flexion arc at all at initial contact. Later in the stance phase, we observed a slight varus deformity of the rearfoot and supination of the plantar sole. Sustained contraction of the TA was visible throughout the gait cycle. The pain was most intense upon heel strike and during loading response and resulted in right-side antalgic gait. The patient walked 10 m in 20.3 sec. On clinical examination, she presented slight edema of the midfoot. Palpation of the distal insertion and last 3 cm of the TA tendon, passive stretching and isometric contraction of the TA and mobilization of the first metatarsal bone elicited pain. This examination also evidenced spasticity in the quadriceps but none in the neurotomized muscles.

A right foot X-ray taken before the patient's consultation in our department did not reveal any abnormality. On bone scintigraphy, early- and late-phase tracer uptake was seen at the right first cuneometatarsal joint (Fig. 1). MRI of the foot revealed distal tendinopathy of the TA, together with interstitial tears, tenosynovitis and edema of the surrounding soft tissues (Fig. 2).

In a single treatment session, BoNT (onabotulinum toxin A, Allergan Inc. Irvine, CA, USA) was injected at 2 points in the right TA (total dose: 70 IU, dilution 1 mL for 25 IU) by an anterior approach and with electrical stimulation guidance. Because TA motor

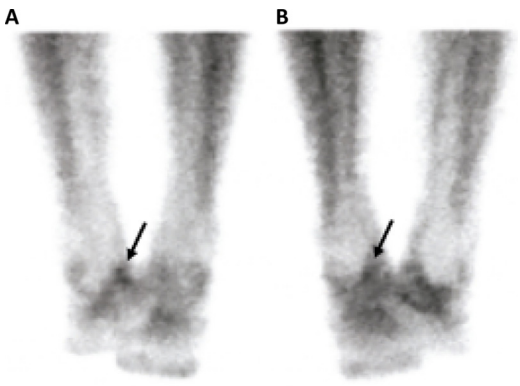

Fig. 1. Bone scintigraphy showing significant tracer uptake (arrow) at the right first cuneometatarsal joint at early (A) and late (B) acquisition times in a 41-year-old woman.

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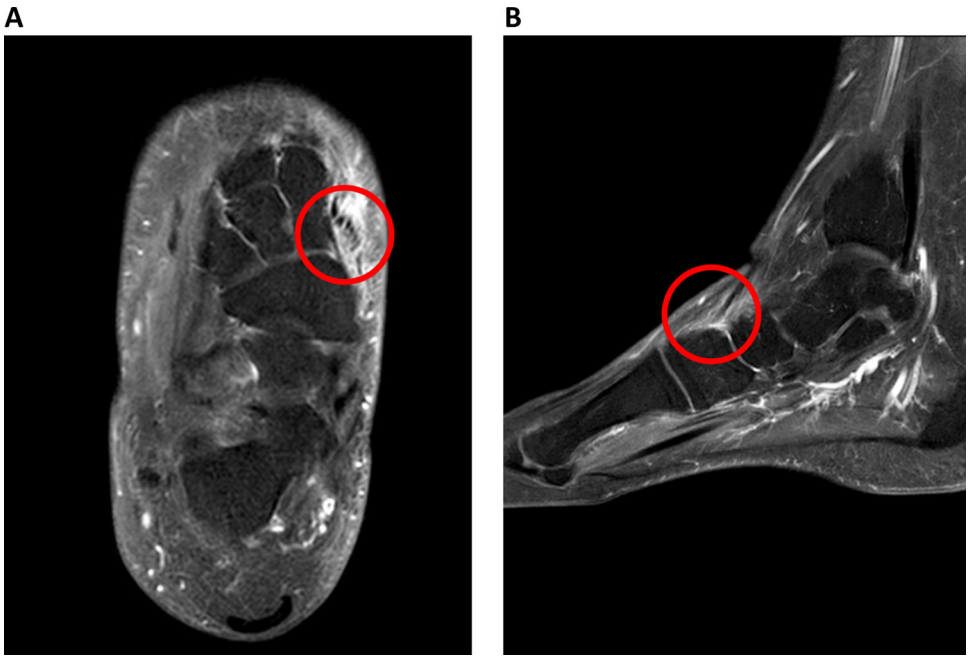

Fig. 2. MRI of the right foot, with transversal (A) and sagittal (B) views. T2 signal hyperintensity (circles) observed in the distal part of the TA tendon, together with swelling of the surrounding soft tissue.

endplates are more highly concentrated in the proximal part of the muscle [1], the first injection was administered a quarter of the way down a line joining the head of the fibula to the external malleolus, and 1 cm external to the tibial crest. The injection was performed in 2 stages (with the needle oriented medially and then laterally, 25 IU in each stage) in order to target fibers located above both sides of the intramuscular aponeurosis. The second injection (20 IU) was administered a third of the way down the above-mentioned reference line, and 1 cm external to the tibial crest.

The treatment goals were to rest the muscle to some extent (allowing the tendon to heal), relieve the midfoot pain, and thus achieve previous levels of gait function and participation in activities of daily living. The frequency of physiotherapy sessions (3 times a week) and focus (deep transverse friction massages and muscle stretching) remained the same after the BoNT injections.

At the 2-month follow-up consultation, the patient reported significantly decreased pain (starting 2 weeks after the injection). The satisfaction index was 9 out of 10. The woman was now able to walk for up to 2 hr (2 km) before being limited by residual pain. Right antalgic gait was no longer present. A first plantar flexion arc was now observed upon initial ground contact. The patient did not complain of drop foot, difficulty in foot clearance during the swing phase, or any other adverse effects. The midfoot edema had disappeared, and stretching and palpation of the TA tendon pain were no longer painful. The patient performed the 10-m walk test in 15.4 sec and had achieved her previous level of participation in activities of daily living. These results were maintained at the 5-month follow-up consultation, although pain level had increased somewhat at 6 months. A second (identical) session of BoNT injection was performed and gave similar results for pain relief and gait function (based on a phone interview). Fig. 3 summarizes the timeline of the case.

Spasticity (i.e., velocity-dependent, passive hypertonia) is often highlighted when considering muscle overactivity after damage to the central nervous system. Lower-limb spasticity usually involves the plantar flexors (causing talipes equinus) and inverters (causing talipes varus). However, muscle overactivity can take other forms [2], some leading to sustained muscle contraction. In particular, dystonic movements occur as involuntary, sustained, patterned muscle contractions of opposing muscles that result in repetitive twisting movements or abnormal posture. In the case we describe, TA dystonia was present at rest but was exacerbated by voluntary activity and gait. Although post-traumatic dystonic movements manifest most frequently as hemidystonia [3], focal dystonia has also been described [4]. The pathophysiology of dystonic movements after head injury has not been characterized, although lesions of the basal ganglia are often mentioned [5]. In the present case, the basal ganglia were not involved. The patient showed white matter damage in the (left) frontal region, which was previously reported in post-trauma patients with dystonia [4].

A variable combination of intrinsic and extrinsic factors most often leads to tendon injury and thus chronic tendinopathy. In the present case, mechanical overload was the main factor because TA dystonia was responsible for long-duration, high-intensity, high-frequency dorsiflexion. In spastic patients, microstructural changes in tendon architecture have been observed [6,7], and some previous studies of hemiplegic shoulder pain have highlighted a relation between pain severity and tendinosis [8,9]. In this context, BoNT treatment led to conflicting results [10], probably because other factors were involved. In dystonic patients, BoNT decreased the pain associated with dystonic postures [11], but, in contrast to our case, the pain was associated with tendon injuries.

The patient's symptoms had not responded sufficiently to well-implemented local physiotherapy. One of the main goals in the treatment of tendinopathy is the relief of mechanical stress on the tendon; however, the sustained nature of the dystonic movement prevented this outcome in our case. BoNT partially paralyzes a muscle, decreases mechanical stress or microtrauma to the tendon, and thus allows healing. Although BoNT constitutes a non-surgical treatment option for lateral epicondylitis (with mixed results [12]), its use to treat tendinopathy at other sites has not previously been reported. We used a moderate dose of BoNT so as to lessen the risk of inducing drop foot. At the 2-month follow-up, the patient reported efficacy in terms of pain but also a positive functional effect on gait (confirmed by an increase in gait speed). The reappearance of a first plantar flexion arc after BoNT injections

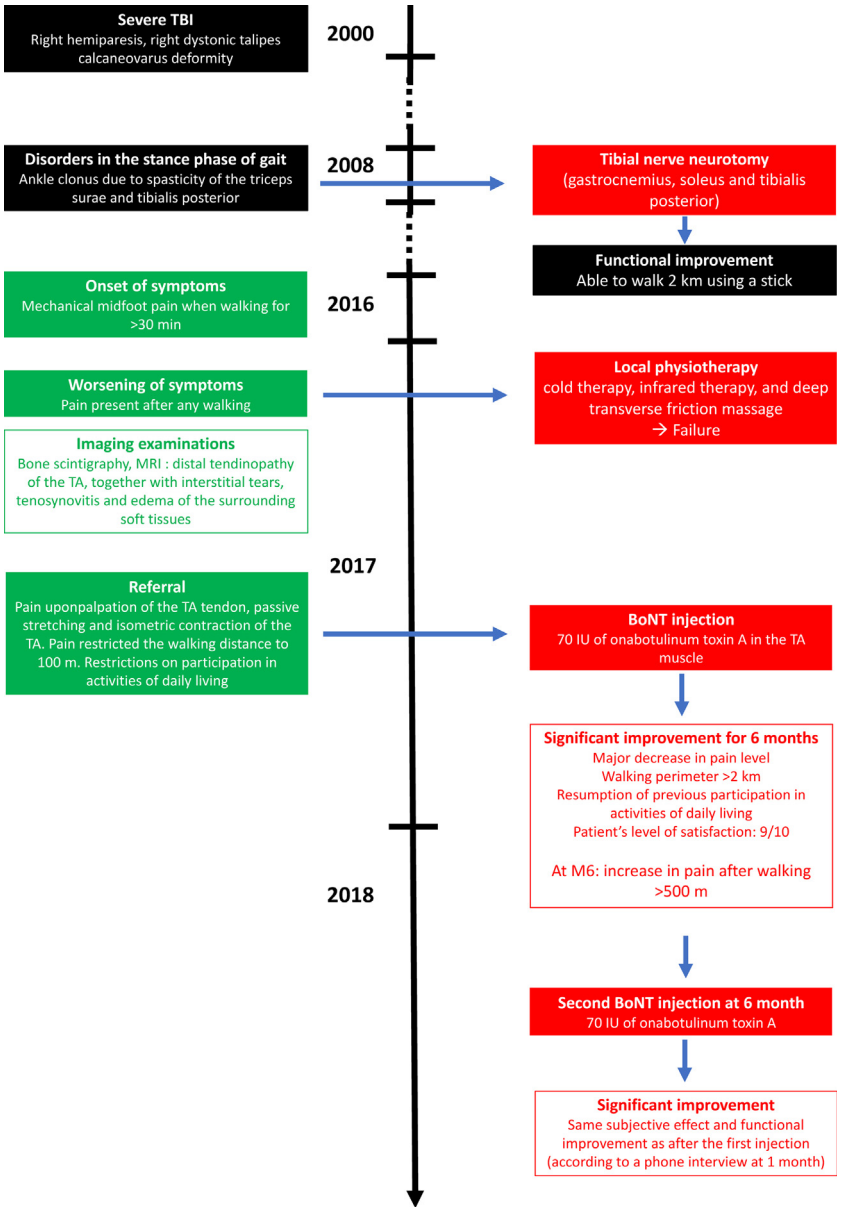

Fig. 3. Timeline of the case. Black boxes refer to the patient's medical history; green elements refer to current illness, physical examinations and diagnoses; red boxes refer to interventions and follow-ups. TBI: traumatic brain injury; TA: tibialis anterior.

might be primarily due to reduced severity of the TA dystonia. However, one could also argue that pain relief could have had a role; eccentric muscle contractions (as performed by the TA at initial contact) are well known to place more stress on the tendon.

The benefits in terms of pain relief and gait function were quite long-lasting (considering the usual period of the BoNT effect), although the treatment had to be repeated after 6 months and the patient will have to be monitored regularly. She still experiences pain when walking long distances but this might be due to residual, non-healed tendon lesions. Although the follow-up period of 7 months was short, our case shows that BoNT may be a valuable treatment option for physiotherapy-refractory tendinopathy in neurological patients and does not induce functional adverse effects. Finally, given that pain was present for the 18 months preceding the BoNT injections, the observed improvement was not likely related to the natural history of the disease.

In conclusion, although severe limb-muscle dystonia is rare after brain injury, it may constitute a risk factor for tendinopathy. As is already true for sports-related tendinopathy, BoNT injection may be safe and efficacious for dystonia-related tendinopathy.

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Disclosure of interest

The authors declare that they have no competing interest.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org.10.1016/j.rehab.2018.06.007.

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