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Partial mixed neuropathy of the fourth lumbar spinal nerve misdiagnosed as “shin splints.”

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A case of anteromedial leg pain diagnosed and treated for 10 years as “shin splints” (medial tibial stress syndrome) is described. A history and examination was performed focused on anatomy, biomechanics, and peripheral nerves. Detailed sensory testing was performed in the painful area, and imaging was obtained to confirm the diagnosis. The clinical investigation was consistent with dynamic stenosis of the left L4-5 intervertebral foramen, causing a mixed partial mononeuropathy of the L4 spinal nerve that presented as pain and hypersensitivity in the anteromedial shin. Manual therapy maneuvers intended to open the intervertebral foramen led to resolution of the pain and sensory deficits. After three additional treatments performed within a month, resolution was maintained for >3 years. This case highlights how concepts from

Neuropathie mixte partielle du quatrième nerf spinal lombaire diagnostiquée à tort comme une “périostite tibiale”.

On décrit un cas de douleur antéro-médiale de la jambe diagnostiquée et traitée pendant 10 ans comme une « périostite tibiale » (syndrome de stress tibial médial). L’anamnèse et l’examen ont porté sur l’anatomie, la biomécanique et les nerfs périphériques. Des tests sensoriels détaillés ont été effectués dans la zone douloureuse et une imagerie a été réalisée pour confirmer le diagnostic. L’examen clinique était compatible avec une sténose dynamique du foramen intervertébral gauche L4-5, provoquant une mononeuropathie partielle mixte du nerf spinal L4 qui s’est manifestée par une douleur et une hypersensibilité dans le tibia antéro-médial. Des manœuvres de thérapie manuelle visant à ouvrir le foramen intervertébral ont permis de résoudre la douleur et les déficits sensoriels. Après trois traitements supplémentaires effectués en l’espace d’un mois, la résolution s’est maintenue pendant trois ans. Ce cas montre comment les concepts issus des études précliniques, associés aux examens

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preclinical studies, coupled with basic anatomical, neurological, and biomechanical investigations, can be critical for accurate diagnosis and treatment for a case previously considered unresponsive to care.

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KEY WORDS: chronic pain, diagnosis, medial tibial stress syndrome, neuropathy, chiropractic

anatomiques, neurologiques et biomécaniques de base, peuvent s'avérer essentiels pour un diagnostic et un traitement précis d'un cas précédemment considéré comme ne répondant pas aux soins.

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MOTS CLÉS : douleur chronique, diagnostic, syndrome de stress tibial médial, neuropathie, chiropratique

Introduction

The term "peripheral neuropathy" has been defined as "a disturbance of function or pathological change in a nerve."[1] A peripheral neuropathy can be of any of the components of the peripheral nervous system, which includes the somatic nerves, the dorsal root ganglia, the dorsal and ventral roots, and the autonomic nerves and ganglia.[2,3] Patients with chronic peripheral neuropathies present clinically with a broad spectrum of symptoms that reflect the affected components of the injured nerve, and can include pain perceived in any structure, altered sensitivities for touch and temperature, and motor and sympathetic dysfunction. Often overlooked is that due to the length of the peripheral nerves, pathologies proximal to the symptoms can cause more distal symptoms. The uniqueness of this case is that the symptoms closely mimicked a medial tibial stress syndrome, while the pathology was 50 to 70 cm proximal to the symptomatic site.

Case presentation

A 23-year-old athletic woman presented with constant pain in her left anteromedial shin and intermittent lower back pain. The symptoms started 10 years previously, following participating in track and field events, particularly the long jump. She was diagnosed and treated as suffering from medial tibial stress syndrome, commonly referred to as "shin splints."[4,5] Medial tibial stress syndrome typically develops following heavy and prolonged exertion, is usually sports-related, and can cause periostitis, stress fractures, and possibly compartment syndrome. The patient had consulted numerous practitioners, including medical physicians, chiropractors, physical therapists, athletic trainers, and a nurse practitioner (Figure 1). X-rays and MRI had been obtained on the left leg and were unremarkable. Extensive treatments had been directed at the shin, and included exercises, deep massage, ice, heat, and electrical stimulation, none of which

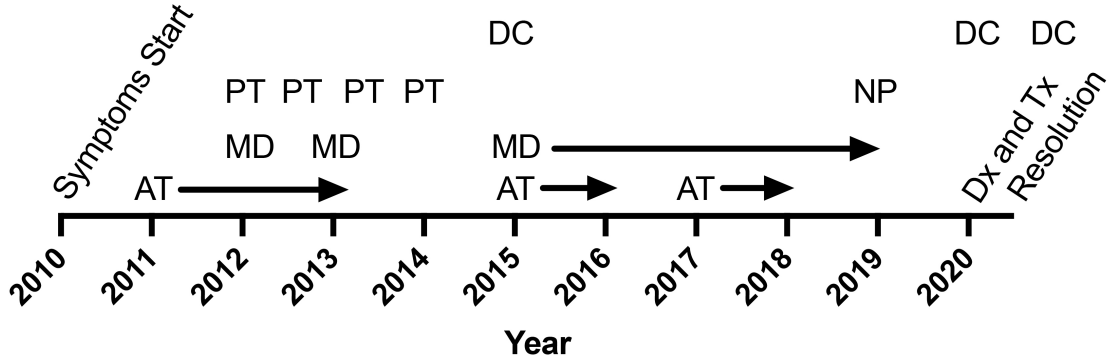

Figure 1.

Timeline of previous care. Each entry indicates a separate provider. AT = Athletic Trainer (High School, 2011-2013; College 2015-2017), MD = Medical Doctor, PT = Physical Therapist, DC = Doctor of Chiropractic, NP = Nurse Practitioner. The third MD cleared athletes for training and competition but did not otherwise provide care. Dx = Diagnosis, Tx = Treatment (by author).

led to more than transient symptomatic changes. The patient had also received care from a chiropractor, but did not mention the shin symptoms. Full-spine X-rays that were read by a radiologist were reported to show "questionable anterolisthesis of L5-S1" but no other structural abnormalities. The patient received 16 treatments that included lumbar spinal manipulation and manual therapy to the neck and upper back.

Over the previous five years, the patient had been a competitive pole vaulter, which had caused the pain to become intermittently more intense, despite continued care as described above. The patient reported sharp pain while in the launching phase of her vaults (when the left hip and lumbar spine were hyperextended). The only time that the patient reported being without pain was during a two-month hiatus from pole vaulting (one year prior to the current presentation). The pain was described as superficial burning and stabbing, and deep aching.

Examination

The patient was exceptionally physically fit and had no postural abnormalities or asymmetries. Lower limb motor power testing reflected the overall high fitness level of the patient and was otherwise unremarkable in that the symptoms were not provoked. There was no tenderness of the symptomatic leg's musculature or of the tibia as would be consistent with periostitis or stress fractures. Because the presenting complaints seemed consistent with a neuropathy of the terminal branch of the saphenous nerve, the entire path of the saphenous nerve was palpated, and was found to be unremarkable for tenderness or reproduction of symptoms. Active lumbar flexion increased back pain, but not leg pain. Active lumbar extension was limited, and provoked the leg symptoms and back pain. Supine unilateral knee and hip flexion (Thomas test) was not painful, but caused the contralateral leg to lift from the table consistent with iliopsoas tightness (bilaterally). Left sided active and passive straight leg raise tests reproduced the shin symptoms at 65° of hip flexion, but not when the hip was externally rotated. When ankle dorsiflexion or hip internal rotation were performed during the straight leg raise at 60° of hip flexion, the shin pain was also reproduced. Performed on the right leg, these tests were unremarkable. Upon prone examination, the paraspinal muscles were symmetrically very dense feeling but not tender, consistent with her high level of fitness, and prevented deeper skeletal structures from being specifically examined. The interspinous ligaments were tender between lumbar (L) and sacral (S) spinous processes of L3 – S1. The sacroiliac joints and gluteal musculature were unremarkable to examination. Prone passive knee flexion with hip extension, designed to traction the femoral nerve, reproduced the shin pain. This test was negative

Figure 2.
Sensory testing of the left anteromedial shin. The reported painful area is outlined. Black dots are where the 0.6 cN monofilament was reported to be painful. At the red dots, neither the 0.04 or 0.6 cN filaments were perceived, but the 2 cN filament was reported to be painful. See text for further details.

when performed on the right. Deep tendon reflexes were normal, there were no strength deficits, and there were no other cutaneous sensory alterations other than described next.

Because the presenting complaints and history seemed consistent with nerve involvement, sensory testing was performed, with the patient unable to see the testing procedure. The painful area was delineated with feedback from the patient using a cotton wisp, by lightly brushing from the surrounding areas until the patient stated that the sensation changed from light touch to pain (Figure 2). Sensory testing was then performed using nylon monofilaments exerting 0.04, 0.6, 2, and 6 cN (Semmes-Weinstein Touch-Test, Stoelting, US). The 0.04 filament was chosen because it was the least force that was consistently perceptible on the entire leg outside the dotted area. The 6 cN filament was the lowest force that evoked a report of being noxious outside the dotted area. At the black dots, the 0.6 cN filament was reported to be painful, and thus allodynic (painful at a stimulus level that was not normally painful). At the red dots, neither the 0.04 or 0.6 cN filaments were perceived (hypoesthesia), but the 2 cN filament was reported to be painful (allodynia).

Imaging

Standing lumbar and thoracic antero-posterior, and lumbar lateral, oblique, flexion, and extension X-rays were performed. The antero-posterior X-ray (Figure 3A) revealed a lumbarized 12th thoracic vertebra (arrowhead, confirmed as T12 with a thoracic view) and a partially sacralized L5 vertebra. The neutral lateral lumbar spine image showed a slightly reduced lordosis (Figure 3B). In extension, the left superior articulating facet of L5 was seen to jut into the L4-5 intervertebral foramen (IVF; Figures 3C and D), and there was a grade 1 retrolisthesis of L4 on L5. MRI of the lumbar spine showed mild narrowing of the left L4-5 IVF (Figure 3E, arrowhead). To determine if there was and pathology affecting the saphenous nerve, it was imaged using ultrasound (from the middle of the thigh to the middle of the shin), and was found to be unremarkable.

Diagnosis

The patient was diagnosed with chronic dynamic inflammatory stenosis of the L4-L5 IVF, discussed in detail below.

Treatment

Two initial treatments were performed as a diagnostic measure, using manual therapy maneuvers biomechanically consistent with opening the involved IVF. The patient was placed prone and the lumbar spine was mobilized in rotation and traction by counter-rotating the pelvis and lumbar spine, using one hand to grasp the anterior ilium while the other palm was placed over the transverse processes of the ipsilateral lumbar spine to stabilize. Each segment was mobilized separately bilaterally using a rhythmic rocking motion. The lumbar spine was then distracted by placing the operator's palm on the sacrum, followed by a cephalad to caudad force, which included both rhythmic oscillations and stronger high velocity but low amplitude pulses. To specifically open the left L4-L5 IVF, the patient was placed on her right side with her torso rotated to the left to the limit of comfortable range of motion, in preparation for side posture spinal manipulation. The operator's thenar eminence was placed on the ileum, and the left hand stabilized the left shoulder of the patient. The operator's right middle finger pulled on the L5 spinous process while the index finger pushed on the L4 spinous process. An impulse was delivered but did not lead to the intended intersegmental movement, which is typically associated with an audible crack or pop. Repeated nerve provocation testing showed that there had been no change. The next day the patient returned, and similar procedures were performed. On this visit however, the side posture manipulation led to a palpable and audible intersegmental movement. The femoral and sciatic nerve testing was performed immediately after this treatment but did not evoke shin symptoms. For two weeks following this treatment, all pain, both ongoing and provoked, were reported to have been absent, after which symptoms reappeared in a less severe form. The patient was referred to a chiropractor for continued care, and received three treatments within a month, similar to the initial treatment with the addition of flexion-distraction technique, also designed to open the IVFs.[6] After each visit, the patient was symptom-free, and after the third visit the symptom relief was maintained. The patient was reevaluated by the author 6 months following the first treatment. Her leg was symptom-free, and she reported one episode of back pain that lasted three days, with an unknown trigger. Sensory examination of the leg was unremarkable, with normal perception thresholds for light touch (0.04

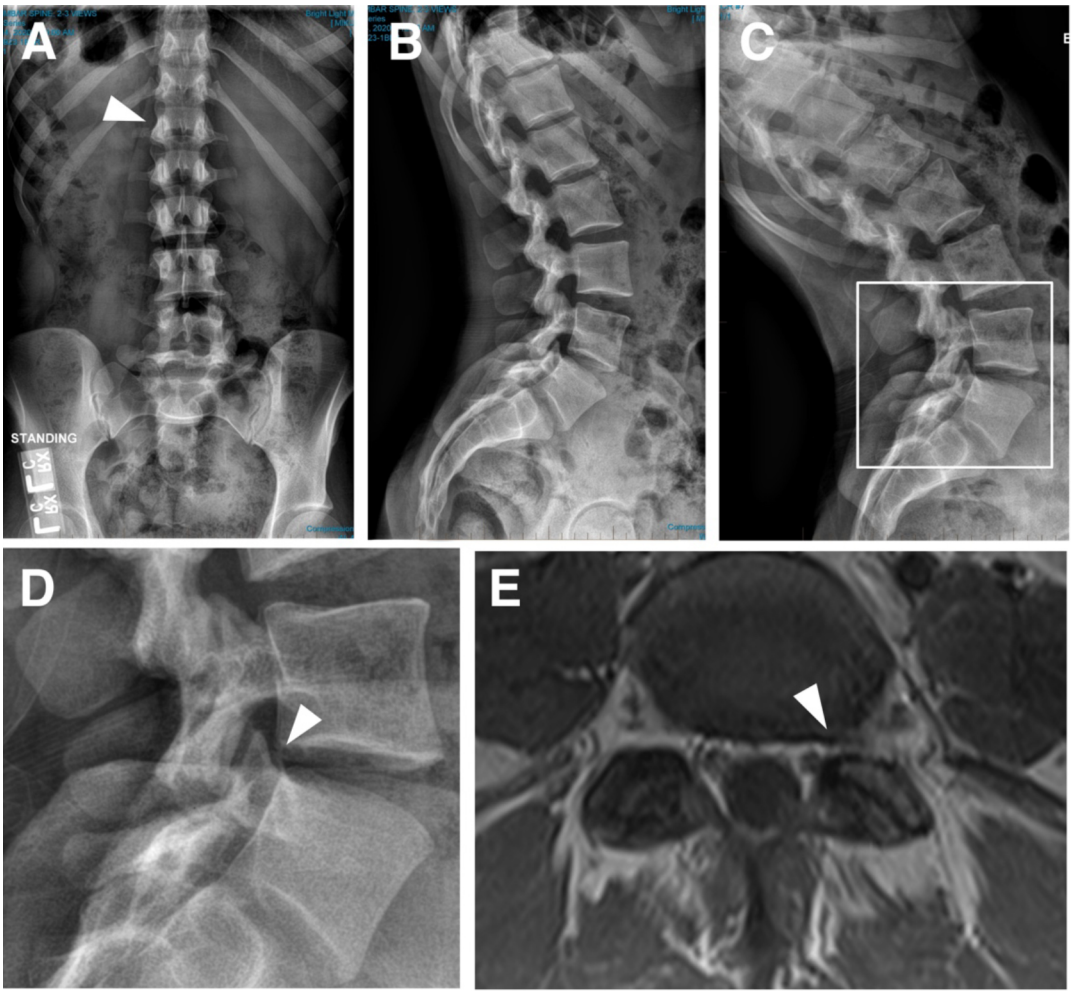

Figure 3.
Imaging of the lumbar spine. A. Anteroposterior lumbar spine, showing lumbarized T12 (arrowhead). B. Lateral lumbar spine. C-D Lumbar extension, showing dynamic stenosis of the L4 intervertebral foramen and retrolisthesis of the L4 vertebra (arrowhead). E. MRI (T1-weighted image) revealed mild static stenosis of the left L4 intervertebral foramen (arrowhead).

cN) and nociception (6 cN) bilaterally. The interspinous ligaments between L4 and S1 remained tender. Lumbar spinal movements were deemed normal. Sciatic and femoral nerve testing did not lead to any symptoms in the leg or in the lower back. At this writing the patient has remained symptom free of the presenting complaint, more than three years after the initial treatment.

Discussion

This patient presented with pain perceived to be deep in the anteromedial leg, and a combination of focal hypo- and hyperesthesias in the territory of the terminal branch of the saphenous nerve. These symptoms are consistent with a mixed sensory neuropathy of the L4 spinal nerve. This initial diagnosis was confirmed using the straight leg raise and the femoral nerve stretch, both of which reproduced the chief complaint of pain in the anteromedial shin. The straight leg raise moves the lumbosacral plexus,[7] which starts at L4. The femoral nerve test tractions the L2-L4 spinal nerves.[8] The only spinal nerve in common with these two tests is L4, thus specifically indicating its involvement. Mild static and dynamic stenosis of the left L4-L5 IVF was confirmed by dynamic plain film X-ray imaging and MRI. While it is commonly held that such mild narrowing of the IVFs is not clinically relevant, this does not consider the possible effects of dynamics of

the motion unit or of inflammation within IVFs, both of which are here posited to be relevant to the clinical presentation.

Pre-clinical observations offer mechanisms for all the painful symptoms suffered by this patient. Passive and repeated hyperextension of the lumbar spine during pole vaulting (the left leg was the launch/stance leg) likely rendered the offending facet joint hypermobile (as seen on the x-ray extension image) and allowed intermittent compression of the L4 spinal nerve. The repeated compression coupled with the excursions of the nerve during normal leg movements[7,9] likely led to inflammation of the nerve[10], known to cause nociceptor axons to fire ectopically and to become sensitive to mechanical and chemical stimulation[11-14]. The paresthesia present at the initial examination was consistent with focal pressure on the L4 spinal nerve and/or axonotmesis. Pre-clinical studies have revealed that nerve inflammation induces ectopic axonal sensitivities.[15,16] Focal nerve inflammation intended to model what may occur in the IVF, induces mechanical sensitivity of nociceptor axons.[11,15,17] This phenomenon is clinically consistent with pain during movements that press or otherwise stress on the nerve, and can occur following focal axoplasmic flow restriction without inflammation.[15,18] Similar research has shown that inflammation also induces nociceptor axons to become sensitive to inflammatory chemicals.[12,19] Clinically this is consistent with pain at rest. Ectopic nociceptor discharge has been shown to induce central sensitization, leading to cutaneous hypersensitivities as documented in this patient.[20] Chronic low levels of nerve inflammation can also lead to signs of axonotmesis consistent with this patient's hypoesthesia, and intraneural inflammation.[21-24] These phenomena have been shown to heal with the resolution of inflammation.[25]

Why this injury only seemed to affect a small part of the L4 spinal nerve is unknown. However, it is unlikely that this is an isolated presentation of a neuropathy, since the presentation of radiating pain syndromes is rarely consistent with segmental or specific nerve distribution patterns.[26] Applying the pre-clinical findings discussed above to cases presenting with radiating pain, including patients with discal herniations, should prove beneficial in the diagnosis, and thus the treatment, of these often-difficult cases.

In this case the single spinal manipulative therapy session immediately relieved the spontaneous and evoked symptoms, a phenomenon that is frequently reported in practice but is not understood. It seems highly unlikely that the presumed inflammation dissipated immediately. However, inflammation is known to cause fibrotic adhesions (scar tissue) in the intervertebral foramina,[27,28] which can tether spinal nerves. Limb movements that call for the spinal nerve to slide, such as hip flexion, would be predicted to transmit abnormal forces to a nerve caught in scar tissue. This could provide the mechanical stimulation to cause the evoked pain and potentially compress the nerve. It is feasible that the manipulation and/or mobilizations disrupted such an adhesion, removing the stimulus even though the inflammation remained. In such a scenario, it would be expected that the adhesion would reform, and that repeated treatments would be required to prevent recurrence, until the inflammation has resolved. In this single case study and with the limitations of current imaging technology it is impossible to verify the proposed mechanism. Interestingly, the immediate (and now maintained) cessation of cutaneous hypersensitivity, likely due to central sensitization, is consistent with previous reports using local anesthetic injections in select cases of neuropathic pain.[29]

Treatments that do not work are often highly diagnostic; in this case, the lack of response to treatments of the shin directed the current practitioner to look more proximally, leading to an accurate diagnosis and treatment approach. In this case, these were indicated by the consultation and physical examination. The plain film findings in extension were important to the final diagnosis. The advanced imaging (MRI and ultrasound) and specialized sensory testing were deemed necessary for this case presentation, but were not necessary for the diagnosis and treatment approach. Finally, it is unknown why the lumbar spinal manipulative treatment that was previously provided did not have a clinical effect on the symptoms, which were reported by the patient to have been severe at that time.

It is unlikely that the treatment permanently resolved or even addressed the mild stenosis or the hypermobility of the spine, leaving the long-term prognosis unknown. The inflammatory process and resulting symptoms could reoccur with resumed intense physical activity, as well as with age-related degenerative changes. The patient was apprised of this likelihood, and was taught exercises

to help stabilize the torso and lower lumbar spine, and movements that may help maintain nerve mobility.

Patient's perspective

Besides the obvious physical pain that I endured for so many years and the setbacks I experienced in my athletic career as a result, the process of seeing different doctors, trainers, and therapists to address my pain led me to immeasurable frustration. I blindly trusted the medical and sports professionals that I saw and they had let me down repeatedly. Upon discovering how straight-forward my condition was and beginning to receive the proper treatment, I experienced not only physical relief from the pain but emotional closure that the pain and complications were not just in my head but were in fact real and, more importantly, could be alleviated and repaired. I am now delighted to be injury free, and I have gotten to experience all over again what it is like to be able to run and move free of pain. I am disappointed that it took so long to get to this point and that I missed out on so many opportunities and unreached potential because of it, but I recognize that this experience has made me a stronger person. I have restored some of my faith in medical professionals but this whole experience taught me the importance of trusting my own body and what I feel instead of what others tell me I should be feeling, as well as getting multiple opinions from professionals before settling.

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