Disappearance of Phantom Limb Pain During Cauda Equina Compression by Spinal Meningioma and Gradual Reactivation After Decompression

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We describe a 65-yr-old woman, whose right lower limb had been amputated at the mid-femoral level because of complicated femur fracture sustained at the age of 5 yr. After amputation, she experienced phantom limb pain (PLP), which gradually decreased in intensity but persisted for 60 yr. At this point the pain diminished progressively, in parallel with the evolution of cauda equina compression caused by an intraspinal tumor. The PLP gradually reappeared over 3 mo after surgical removal of the tumor.

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Phantom limb pain (PLP) is a pain that may be seen immediately after injury or years later in a part of the body that is deafferented or amputated. It is seen in 60%–80% of patients after amputation and is described as being intermittent or persistent cramping, stabbing, squeezing, aching, burning, and shooting (1). Although PLP may remain constant for years during the postamputation period, both the frequency and the duration of pain attacks decrease significantly (2).The exact causes underlying PLP remain unknown. However, studies have demonstrated that PLP is a complex phenomenon that may be attributed to neural reorganization at multiple levels, including the spinal cord, brainstem, thalamus, and cortex, as well as factors in the periphery (3).

This is the first reported case, in which PLP disappeared progressively, paralleling the evolution of cauda equine compression because of an intraspinal tumor and gradually reappeared on slow recovery from cauda equina syndrome 3 mo after surgical removal of the tumor.

Case Report

A 65-yr-old woman was scheduled to undergo surgery for cauda equina syndrome resulting from an intraspinal tumor. Her lower right leg had been amputated at the mid-femoral level because of complications from a femur fracture when she was 5 yr old. After amputation, she suffered from an intermittent, electric shock-like pain that radiated from her buttock to below the level of her amputation and into her phantom foot and toes. The pain was triggered by cold and was decreased by standard analgesics. Over time, her pain gradually decreased in intensity but persisted for 60 yr. The patient was admitted to hospital with the complaint of motor weakness and sensory loss in the left leg and urinary incontinence. Approximately 1 yr before admission to the hospital, paresis of the lower extremities had begun and the old pain in her amputated leg had gradually decreased and disappeared. She experienced urinary incontinence just before hospital admission. At neurological examination, paraparesis and hypoesthesia were observed below the level of the second lumbar segment. Magnetic resonance imaging (MRI) revealed a calcified and hyperintense intradural mass at the level of L1-2 compressing the cauda equina (Fig. 1). Spinal intradural meningioma was diagnosed, and a surgical decompression with laminectomy was performed. The mass, which totally obliterated the spinal canal, was removed completely. Its histopathological diagnosis was psammomatous meningioma. When the patient was discharged 1 wk after operation, dysesthesia in her left leg and saddle region showed some improvement, but she was still complaining of inability to urinate and left leg weakness. Full sensory function and sphincter control recovered approximately 3 mo later, but she began to complain of an intermittent sharp shooting and burning pain that was characteristically originating in the distal area of the amputated leg and radiated into her phantom foot, just like she had

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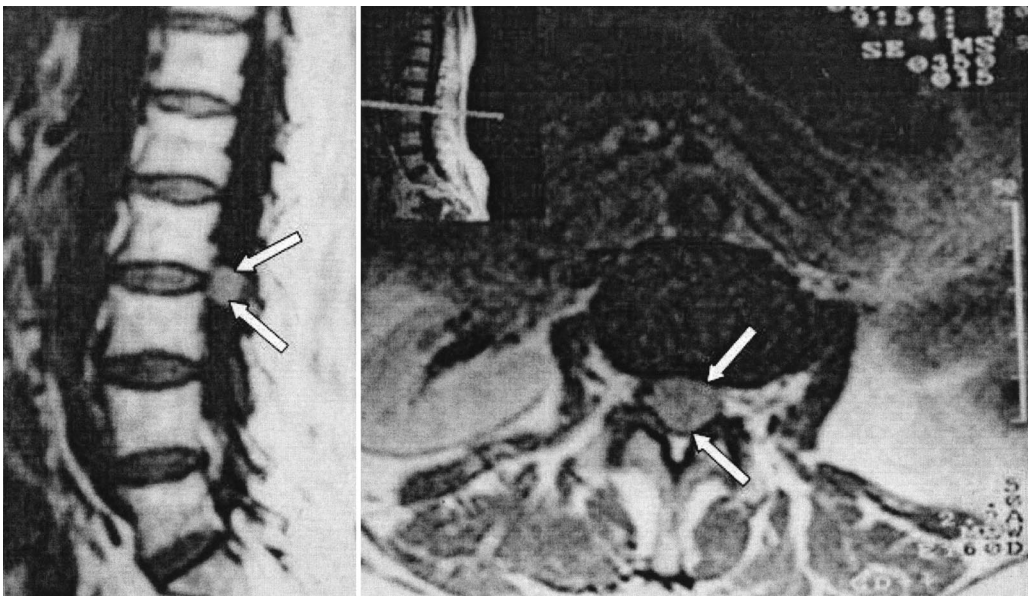

Figure 1. Magnetic resonance imaging (MRI) of the lumbosacral spine. The sagittal (left) and axial (right) views demonstrate a calcified and high signal intensity intradural mass (arrows) at the level of the L1-2, which obliterated the spinal canal

before. Any neuroma formation, undue osseous prominences, or scar tenderness could not be detected in the stump. Her pain was reduced with oral fluoxetine (20 mg per day), which she had previously taken. At the 10-mo follow-up evaluation, MRI showed no lesion in the lumbar spinal canal (Fig. 2) and the patient was completely free of all neurological symptoms of cauda equina syndrome; however, her phantom pain had reappeared.

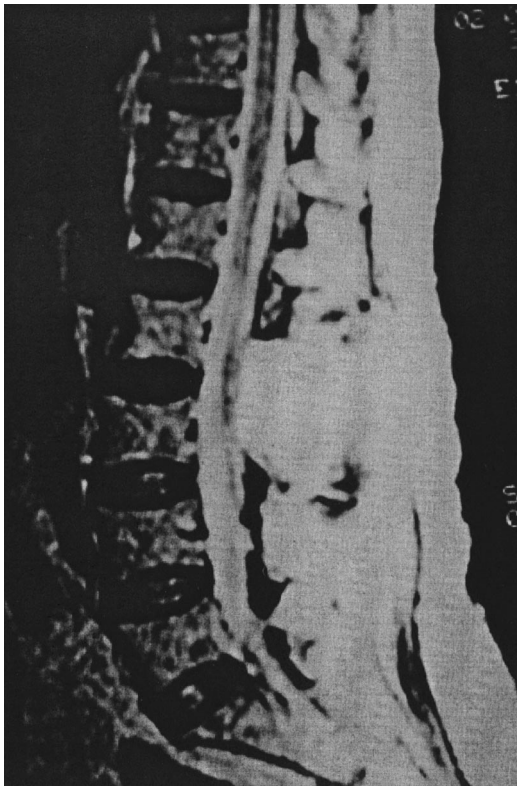

Figure 2. Magnetic resonance imaging showed no lesion in the lumbar spinal canal 10 mo after the operation.

Discussion

Our patient's PLP had disappeared with the beginning symptoms of a cauda equina syndrome caused by meningioma and gradually reactivated after surgical removal of the tumor. We thought that the suppression of the PLP was caused by the mass-induced ischemic and mechanical damage to the cauda equina and that its reactivation was caused by electrophysiological improvement after removal of the mass. During cauda equina compression, nerve fibers have differing susceptibilities both to ischemia and mechanical trauma. The small myelinated and unmyelinated fibers (e.g., Aδ and C-fibers) that subserve pain sensation and parasympathetic function are much less resilient to mechanical compression than the larger fibers concerned with motor power, light touch, and joint position sense. Reversibility is dependent on relief of both the mechanical pressure obstructing axoplasmic flow and the resolution of ischemia/venous congestion (4).

The mechanism of mechanical damage is used clinically for the treatment of trigeminal neuralgia in the balloon compression technique. Balloon compression of the trigeminal nerve has been used effectively to treat trigeminal neuralgia for two decades. The compression injures large myelinated fibers, removing the trigger to the presumed ephaptic transmission of pain. Because small unmyelinated fibers, which mediate the

corneal reflex, are selectively preserved, the compression technique may be an advantage in the treatment of ophthalmic division nerve pain (5).

Some studies demonstrated that evoked potentials in the medulla spinalis suppressed by cervical medullary compression recovered promptly after surgical decompression (6). Brihaye (7) reported a case in which painless phantom phenomenon extinguished itself progressively, paralleling the aggravation of cervical radiculopathy because of a herniated disk and reappeared on slow recovery from radiculopathy 6 months after removal of the herniated disk. Iida et al. (8) reported another similar case, in which PLP was relieved and remained asymptomatic for 15 years reactivated after cervical spinal decompression.

Conversely, secondary phantom limb problems, reactivated after an asymptomatic interval after the amputation, have been reported after intervertebral disk hernia, spinal cord injury, and vertebral neoplasia (9–12). Pithwa and Rajasekaran (13) reported an interesting case in which secondary PLP developed as a result of a cervical epidural tubercular abscess and was relieved after treatment with antituberculous therapy.

This phenomenon may be dependent on whether the individual nerve fibers are partly or completely affected and whether the cauda equina or medulla spinalis is incompletely or completely compressed. Partial loss of cauda equina or medulla spinalis functions therefore may cause little or no disability (14). For reactivating PLP just after decompression, another possible explanation may be that mechanical stimuli or inflammatory responses resulting from the surgical stress to the spinal cord resulted in functional changes in the afferent pathways of the amputated limb. But, in our case and Brihaye's (7) case PLP was reactivated 3 and 6 months, respectively, after decompression. Therefore, this mechanism is excluded in our case.

Regional anesthesia with local anesthetics provides a reversible and transient deafferentation model in humans and may be hypothesized as sharing fundamentally similar mechanisms. Many cases of reactivation of PLP in amputees have been reported during neuraxial, plexus, and peripheral nerve blocks (15–17).These examples support the theory, elaborated by Melzack (18), that this is attributable to a decrease in the tonic inhibitory influence exerted by the brainstem reticular formation, permitting pain. However, Tessler and Kleiman (19) prospectively studied 23 spinal anesthetics in 17 patients with previous lower limb amputation. Only one patient developed clinically significant PLP. The difference may depend on whether conduction of the individual nerve fibers, plexus, or spinal cord ceased completely or partially.

Martin et al. (15) presented a case in which lumbar plexus block resulted in apparent unmasking of PLP previously experienced in the distribution of the sciatic nerve, and PLP was subsequently alleviated by sciatic blockade. They suggested that afferent input from intact axons in neighboring peripheral nerves (which share the same spinal cord segments) may exert an influence in the tonic inhibition of phantom pain generated peripherally by injured axons.

Paqueron et al. (20) described a late-onset PLP during a continuous analgesic popliteal nerve block with small-concentration local anesthetic after foot surgery and its alleviation and recurrence when stopping and resuming the local anesthetic infusion. They suggested that small-concentration local anesthetics block the small- diameter fibers (Aδ and C-fibers,), and a partial deafferentation of these small sensory fibers might lead to PLP.

In summary, we present a case in which the PLP in the amputated leg disappeared during cauda equina compression by meningioma and reactivated after surgical decompression. This case suggests that although incomplete compression or block with local anesthetics may result in reactivation of PLP that had once been completely relieved and remained asymptomatic for years, complete compression or blockade of nerves, a nerve plexus, cauda equina, or medullary cord may result in suppression of PLP, and decompression or recovery of the block may cause reactivation. In conclusion, the cited literature and the present case highlight the relative importance of peripheral and central mechanisms in PLP.

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