Coupled obturator neurotomies and lidocaine intrathecal infusion to treat bilateral adductor spasticity and drug-refractory pain

Case report

JOSÉ D. CARRILLO-RUIZ, M.D., PH.D.,1,2 PABLO ANDRADE, M.D.,1,2 NORA GODÍNEZ-CUBILLOS, M.D.,3 MARÍA L. MONTES-CASTILLO, M.D.,4 FIACRO JIMÉNEZ, M.D., PH.D.,1,2 ANA L. VELASCO, M.D., PH.D.,1 GUILLERMO CASTRO, M.D., M.SC.,1 AND FRANCISCO VELASCO, M.D.1

1Functional Neurosurgery, Stereotaxy, and Radiosurgery Unit, Mexico General Hospital; 2Anáhuac University, School of Medicine and Psychology; 3Algology Unit, Mexico General Hospital; and 4Rehabilitation Unit, Mexico General Hospital, Mexico City, Mexico

Spastic diplegia is present in three-fourths of children with cerebral palsy, interfering with gait and frequently accompanied by severe pain. The authors report the case of a 28-year-old woman with history of perinatal hypoxia, who presented with cerebral palsy and severe spastic diplegia (Ashworth Scale Score 4, Tardieu Scale Score 5) and was confined to a wheelchair. She complained of pain in the left hip and knee with mixed neuropathic and somatic components. She consistently rated pain intensity as 10 of 10 on a visual analog scale, and her symptoms were resistant to multiple treatments. The patient underwent selective bilateral adductor myotomies and the implantation of an infusion pump for intrathecal lidocaine application. Postoperative control of pain and spasticity was dramatic (scores of 0 on the Ashworth, Tardieu, and visual analog scales) and persisted throughout a follow-up period of 36 months. This is the first report in the literature of combined selective neurotomies for the treatment of spasticity and chronic lidocaine subarachnoid infusion to treat associated pain. This therapy could represent an alternative to treat spasticity associated with neuropathic and somatic pain. (DOI: 10.3171/2009.11.JNS09607)

Key Words • spasticity • neuropathic pain • cerebral palsy • diplegia • infusion pump • neurotomies

Spasticity is an abnormal increase in muscle tone due to neurological insult that can lead to permanent muscle contracture, painful spasms, clonus, and lack of voluntary movements of extremities. Cerebral palsy is the most common cause of spasticity in childhood, and approximately 75–80% of patients with cerebral palsy present with a certain type of spasticity. Spastic diplegia is frequently accompanied by a scissor pattern that interferes with gait and is often associated with disabling somatic pain. Occasionally, long-term elongation of nerves caused by extreme diplegic postures may cause nerve fiber lesions in the hip articular capsule or trapped nerves in the crural area, both resulting in neuropathic pain.

Abbreviation used in this paper: VAS = visual analog scale.

Traditional treatment for spasticity includes a combination of physical therapies, motor point blocks, drug therapies, and surgical procedures. Selective microsurgical neurotomies are effective at controlling focal spasticity. Infusion pump implantation for delivering subarachnoid drugs has been used to treat regional spasticity. Baclofen is the most accepted drug for this purpose. The most frequent complication associated with baclofen usage is the withdrawal syndrome. However, due to the high cost of the drug, the fact that baclofen is unavailable in many countries, and the serious consequences associated with its withdrawal, other medications have been investigated. Additionally, baclofen is highly effective at controlling spasticity but less so at alleviating pain.

Previous experiments in animal models have suggested that systemic infusion of lidocaine relieves neuro-
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pathic pain, but to our knowledge, there are no reports of the combination of neurotomy and intrathecal lidocaine infusion to treat spasticity and pain. The objective of this article is to describe the techniques involved in combining neurotomies for spasticity and intrathecal lidocaine infusion for pain.

Case Report

History and Examination. This 28-year-old woman had a history of perinatal hypoxia accompanied by cerebral palsy, mild mental retardation, and a torpid psychomotor development. During childhood, a bilateral adductor myotomy was performed by the orthopedic service. Nevertheless, the spasticity returned a few years later in the form of severe spastic diplegia. At physical examination the patient presented with scissor gait that kept her from walking, and she was confined to a wheelchair.

She also exhibited severe spasticity in both thighs and arthrodesis of the left knee as a result of corrective surgery. Spasticity scores were 4 and 5 on the modified Ashworth and Tardieu scales, respectively. The patient also had referred neuropathic and somatic pain in the left hip and knee, and a 10-point VAS was administered. Neuropathic pain symptoms included paresthesias over the inguinal region and hyperalgesia with burning pain sensation in this area. Somatic pain was present even at rest. However, when the hip and leg moved either by spontaneous spasm or passive movement, the VAS score reached 10. This completely reduced the opportunity of starting rehabilitation therapy. Orthopedic evaluation reported femoral head impacted into the pelvic acetabulum of the left hip during spasms, which caused intense pain. The goal of the surgical procedure was to treat spasticity as the best option for this patient.

Operation and Postoperative Course. After administration of an epidural blockade and a local anesthetic, we placed the patient in the dorsal decubitus gynecological position. The surgical approach included an inguinal incision in each side, taking as reference the ischium and pubic bones. The soft tissue was identified and opened. The adductor muscles and their tendons were identified, and dissection was performed between the adductor fascicles to expose the obturator nerves. Guided by electrostimulation of the nerve fascicles, we conducted selective total motor neurotomies of the anterior and posterior obturators bilaterally, as well as myotomies and tenotomies of the adductors. Progressive dissection and cutting of the obturator fibers, while testing with Ashworth and Tardieu scales, determined when spasticity of the involved muscles was relieved. The postoperative course was uneventful. In the immediate follow-up period, the patient’s spasticity was significantly diminished, but pain persisted mainly in the anterolateral aspect of the left lower limb. The patient was treated with various analgesics and multiple peripheral nerve blocks (femorocutaneous, femoral, and pudendal in a technique called “three-in-one”), which resulted in transient modest analgesic effect; however, the therapy had to be repeated every 3 months and was carried on up to 1 year.

In a second procedure, 12 months after the neurotomies, the patient underwent surgery for intrathecal catheter implantation, leaving the tip at the T-7 level connected to an external infusion pump (Baxter Infusion Pump [65 ml]). Lidocaine 2% was administered at 0.3 ml/day for 1 month. Due to a dramatic decrease in pain intensity, we considered implanting a continuous infusion pump. At 13 months, the epidural catheter was changed for a definitive subarachnoid one and connected to an Isomed Implantable Infusion Pump (Model 8472–35–10, Medtronic, Inc.). The pump was filled with 35 ml of lidocaine (2%) to fill the reservoir and the dosage was administered at the regular constant pump rate.

Postoperative radiological images showed the correct location of the intrathecal catheter. The pump was recharged every 3–4 months (Fig. 1). Clinical evaluations with the same presurgical scales were performed at 0, 3, 6, 9, 12, 24, 30, and 36 months to test spasticity and pain (Fig. 2). The patient’s spasticity gradually improved to a functional level. She became able to stand and ambulate with help and started physical therapy. Pain control was highly effective after implantation of the intrathecal lidocaine pump and remained so until her last evaluation 36 months after operation. At this time, all analgesics had been discontinued. She repeatedly reported a VAS score of 0.

Discussion

We have presented the case of a patient with cerebral palsy treated for spasticity and pain with a combination of methods. Obviously, it would have been desirable to build up a series of cases to strengthen the scientific merit of this report.

In our case, spasticity, considered as a pure motor illness, was accompanied by severe pain, probably secondary to joint deformation with extreme adduction and hip dislocation, which were increased during spasms, corresponding to somatic or muscle pain. The forced position of the extremities had probably caused chronic compression of inguinal-region nerves, which generated dysesthesias.

Fig. 1. A: Identification of the obturator nerve after dissection of the adductors. B: Nerve electrodes were used during surgery to identify adductor muscle contractions and prevent sectioning of sensitive fascicles. C: Postoperative radiological image showing the correct location of the infusion pump.
and hyperalgesia of the inguinal and crural area and may be considered neuropathic pain.

Since the early years of functional neurosurgery, obturator neurotomies have been performed in the treatment of spasticity. In our case, elective neurotomies resulted in relief of adductor muscle spasticity, which allowed abduction of the lower extremities, avoidance of the scissor posture, and improved gait.

Although spasticity relief was evident in the immediate postoperative period, our patient continued to suffer severe left hip and crural area pain, which prevented her from initiating rehabilitation therapy.

The use of lidocaine to treat pain is an old treatment and local anesthetic. Successful sensory block has been previously described after subarachnoid lidocaine infusion in experimental animals, demonstrating that it has a local effect on the subarachnoid structures. A unique study in the literature reported a single case in which the application of lidocaine at the spine exacerbated the neuropathic pain scores and required immediate morphine application to diminish pain. The long-term therapy proved to be efficacious in preventing the recurrence of pain. At the time of the present study of acute intrathecal lidocaine administration, there is nothing in the literature on chronic intrathecal lidocaine therapy for the treatment of pain.

There have been reports of side effects when using lidocaine in spinal blocks, occurring in 0.01–0.7% of cases. Side effects include paresthesias, perineal dysesthesias, and sphincter dysfunction with transient muscular weakness in the extremities. However, these effects are dose and concentration dependent. In our case, the administration of intrathecal lidocaine 0.3 ml/day in a 2% concentration was not accompanied by any side effect for over 2 years.

Although lidocaine’s mechanism of action is still not well understood, it is believed to act over the voltage-gated sodium channels. Restriction of the electrolyte flow through these pores prevents development of action potentials in neurons and muscles. The anesthetic effect results from hyperpolarization of membranes, which interferes with nociceptive signals.

Our case demonstrates the convenience of combining lesional and modulatory techniques; coupling spasticity surgery with intrathecal lidocaine infusion to treat pain yielded a very positive synergic result. The surgical effect on spasticity resulted in early improvement that was maintained throughout the follow-up period. However, nerve blocks with lidocaine as a diagnostic test to anticipate the effect of selective neurotomy for spasticity produced only transient improvement of pain. In contrast, long-term intrathecal lidocaine administration resulted in effective control of somatic and neuropathic pain.

Low-dose intrathecal lidocaine was considered safe. It is also inexpensive and readily available for pump refilling. This seems important in countries where baclofen is not available or is too expensive.

**Disclosure**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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References


