Unilateral Lumbosacral Plexopathy Following Extra-Corporeal Membrane Oxygenation Managed with Burst Spinal Cord Stimulation: A Case Report

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Background: Lumbar plexopathy is considered a rare complication of minimally invasive endovascular or surgical procedures, but it has not been related to the use of extracorporeal membrane oxygenation (ECMO) so far. In cases of high-intensity neuropathic pain, neuromodulation should be considered early in the treatment.

Case Report: After receiving supportive ECMO therapy for 7 days, a 42-year-old woman developed lumbar plexopathy and high-intensity neuropathic pain and was referred to the pain unit 6 months later. After unsuccessful use of neuropathic drugs, spinal cord stimulation achieved global improvements of 90%. Sixteen months later, the patient continues to be asymptomatic, leading an active life as a mother of 4 children.

Conclusions: The use of ECMO cannot be said to be the main cause of plexopathy in this case, but a multifactorial approach regarding this issue should be considered. Spinal cord stimulation can provide dramatic relief in localized neuropathic pain.

Key words: Extracorporeal membrane oxygenation (ECMO), lumbosacral plexopathy, spinal cord stimulation

BACKGROUND

Lumbosacral plexopathy (LSP) has a multifactorial etiologic origin; its incidence is low and can only be diagnosed indirectly. Approximately 20% of these patients develop femoral neuropathy (1).

If the origin is traumatic or ischemic, prognosis is often poor. Some case series in the literature describe spontaneous improvement in about two-thirds of the cases after 18 months, while 9% became chronic LSP (2).

To understand the potential etiological mechanisms involved in its genesis, a thorough anatomic knowledge of these structures is mandatory (Figs. 1 and 2).

The lumbar plexus consists of anastomoses between the anterior rami of the first 3 lumbar nerves (L1-L3) and a portion of the fourth lumbar nerve (L4).

The sacral plexus is formed by the union of the lumbosacral trunk (L4-L5) with the anterior rami of the first sacral nerves (S1-S3) (3).

Vascularization of the lumbosacral plexus is achieved by the 5 lumbar branches of the abdominal aortic artery, deep circumflex iliac artery, and branches of the gluteal and internal gluteal arteries, as well as muscular branches of the psoas (4,5).

Lesions have been noted after aortoiliac therapeutic procedures, both in minimally invasive endovascular interventions and in major surgeries (6-8).

The ischemic origin of LSP is usually associated with

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kidney transplants and multiple trauma patients who recovered after prolonged hypovolemia and cardiac arrest. In these cases, the common denominator is the low cardiac output induced by the graft’s blood theft (steal syndrome) due to hypovolemic shock (9).

Extracorporeal membrane oxygenation (ECMO) constitutes an advanced life support system during lung or cardiac failure. No case of unilateral LSP related to ECMO has been described in the literature, but there is one case of bilateral LSP and hemiplegia persisting after 18 months (6).

The lumbosacral plexus has both motor and sensitive branches. Clinical attention is often focused on the motor deficit, although the generated neuropathic pain (NP) is very intense and affects quality of life and catastrophism dramatically, reducing the chances of recovery (7,11). Its main mechanisms are deafferentation, pathologic compressive stimulation of nerve fibers, and sympathetically mediated activation (12,13).

The femoral nerve is the main branch of the lumbar plexus, so femoral neuropathy is characterized by weakness in the femoral quadriceps, decrease and/or loss of the knee-jerk reflex, and decrease or loss of sensitivity in the anterior medial part of the thigh and anterior distal part of the leg, sometimes accompanied by paresthesia. In cases of retroperitoneal hematoma (RPH) or direct trauma, the initial symptom is the appearance of severe pain referred to the anterior and medial thigh. Then, motor dysfunction progressively develops. If the cause is not addressed, muscle weakness and atrophy appear (14).

Similarly, motor and sensory symptoms in the posterior thigh, leg, foot, and perineum may suggest the presence of affected nerves in the sacral plexus, whose main branch is the sciatic nerve.

Early LSP diagnosis should be performed using a polyneuropathy diagnostic protocol, including detailed clinical examination, complete analytics, and immunologic blood and cerebrospinal fluid studies if the etiology is unclear. Neurophysiological assessment should include
Spinal cord stimulation (SCS) is a valid option for the long-term relief of trunk and limb neuropathic chronic pain (16). Although a case report describing the management of a patient suffering from LSP secondary to pelvic trauma/surgery was published in 2005 (17), as far as we know there are no reports on the application of SCS in patients with LSP following ECMO.

CASE

A 40-year-old woman with no relevant medical history, apart from the episode described here, was referred to our unit from the neurology service with refractory NP. The patient arrived at the emergency department with right hemicrania and midthoracic oppressive sensation, dizziness, general unrest, and vegetative courtship. The electrocardiogram revealed ST elevation at DI, aVL, and V1-V6 with bigeminism, frequent extrasystoles, signs of hypoperfusion, and marked hypotension. An emergency catheterization showed normal coronary arteries. Echocardiography revealed a severely depressed left ventricular ejection fraction (LVEF), mild left-ventricle hypertrophy, and hyperechogenic appearance, suggestive of myocarditis. The patient was admitted to the intensive care unit (ICU) with atrial fibrillation of 140 beats per minute, significant electric lability, phases of bigeminy, spells of nonsustained ventricular tachycardia, left bundle branch block, and signs of poor peripheral perfusion. Thus, she was treated with vasoactive medication, but clinical worsening of symptoms with poor echocardiographic evolution and severe systolic dysfunction (LVEF 15%) during the first hours led to the implantation of an ECMO system. Immediately after ECMO removal, the patient experienced pain from the upper right buttock, anterolateral thigh toward the knee, and irradiation along the internal side of the leg down to the ankle, with sensitive alteration and motor weakness, which made ambulation impossible. Hours later, pain and motor alterations in the femoral area worsened and had to be controlled using morphic rescues. Cranial and abdominopelvic CT, doppler echocardiography, and MRI failed to identify any cause that could justify the patient’s clinical situation.

Fentanyl patches (25 µg/72 h), pregabalin 75 mg/72 h, and on-demand intravenous morphic rescues plus first-step analgesia achieved better control relief. As all imaging tests were negative and analgesic doses were increased during the first days, a comparative neurophysiologic study in both lower limbs was performed. The results suggested right lumbosacral plexopathy with sensitivity predominantly in the sural, superficial peroneal, and internal saphenous nerves, as well as femoral nerve motor impairment.

After being diagnosed with lumbosacral plexopathy and having recovered satisfactorily from the cardiogenic shock, the patient was discharged with a fentanyl patch of 37.5 µg/h for 72 hours and pregabalin 150 mg/12 h. Cardiologic evaluation revealed ejection fraction improvement up to 75%.

Twelve months after discharge, the patient visited our pain unit with pain in her right lower limb, radiating to the buttock area, anterior and lateral thigh, and anterolateral leg down to the ankle, in addition to allodynia in the whole thigh, hyperalgesia in the right crural area, and quadriceps weakness at 4 of 5 with a preserved osteotendinous reflex. Autonomous walking without technical aid and muscular balance exploration was impossible due to pain.

The patient’s Visual Analog Scale (VAS) score was close to 10, with peaks of irruptive pain, presenting neuropathic characteristics with significant scores on the Leeds Assessment of Neuropathic Symptoms and Signs (LANSS), Douleur Neuropathique 4 Questions (DN4), and PainDetect scales which, combined with a mixed anxiety-depressive disorder, prevented the patient from any work or family and household activities.
A new neurophysiological study confirmed the diagnosis of right lumbosacral plexopathy with predominant sensitive sural, superficial peroneal, saphenous, and femoral affection (15). Basic neurological examination revealed allodynia and hyperalgesia in the affected area. In addition, the pathological QST of the temperature threshold showed positive results (18).

We performed a trial of spinal cord stimulation (SCS) and implanted 2 octapolar leads (Octrode®, Abbott, Plano, TX) in the epidural space midline at the level of T8, achieving paresthesia in the painful area.

During the SCS trial, the patient reported improvements of approximately 60% with traditional tonic stimulation and higher than 90% with burst stimulation, sustained over 21 days. Thereafter, a permanent pulse generator (Prodigy MRI®, Abbott, Plano, TX) was implanted in her right upper buttock. Burst stimulation did not produce paresthesia, thereby reducing discomfort. It was programmed in trains of five 1000 µs/0.5 mA pulses with 500-Hz intraburst and 40-Hz interburst rates, in cycles of 30 seconds on/90 seconds off (19,20).

Burst stimulation mimics the activation patterns of nociceptive pathways. There are 2 afferent pathways and one inhibitory pathway for nociceptive stimuli. The medial ascending pathway encodes the motivational/affective component of pain (unpleasant sensation). It is activated by C-fibers and connects to the medial dorsal and ventral posterolateral nuclei of the thalamus (20).

At 16 months postoperatively, symptomatic improvement persisted with the total withdrawal of opioids and antineuropathic medication. Her VAS score was 20 mm on a 100-mm scale, with a substantial increase in her quality of life and work capability. The patient declares herself to be very satisfied with the therapy. No adverse events related to the therapy have been observed.

**DISCUSSION**

LSP lesions during interventional procedures may be due to nerve trauma by direct compression, hematoma, or pseudoaneurysm formation. Indirect ischemic lesions secondary to the use of percutaneous closure devices, surgical retractors (21), or suturing (22,23) have also been reported. The most frequent complications of ECMO devices are vascular. The incidence of acute ischemia in the lower limbs ranges from 10% to 70%. Other described adverse events, such as arterial dissection, pseudoaneurysm, and RPH (24,25), may be potential causes of nerve lesions. Patients most likely to develop this kind of complication are those with preexisting arterial disease and young women who have not developed collateral circulation as they did not have a previous arteriosclerotic chronic disease (26,27). Large cannulas used for drainage and return may compromise blood flow in the distal area of the device. Therefore, placing a retrograde mechanical bypass decreases the incidence of acute ischemia and is recommended wherever possible (28).

In patients with refractory NP and low quality of life, SCS is a valid treatment option and has been recommended by the National Institute for Health and Care Excellence. According to these recommendations, a successful SCS trial (pain relief > 50%) is mandatory before permanent implantation, and care should be provided in a multidisciplinary manner (29).

While “classic” low-frequency paresthesia-based SCS has high possibilities of obtaining 50% pain relief, other newer stimulation modalities and targets such as high-frequency and burst, as well as the stimulation of the dorsal root ganglia, increase the chances of achieving higher sustained relief (30).

In our case, in the absence of RPH identifiable by image scanning, we considered that the neurological damage most likely had an ischemic origin. It is induced by a double mechanism: severe cardiogenic shock with low cardiac output, and high doses of vasoactive drugs with large-diameter cannulas in the femoral territory in a young female patient without collateral circulation (31,32).

In a recent and comprehensive American publication that reviewed 15,894,201 patients undergoing femoral percutaneous catheter procedures, the incidence of femoral nerve lesions was 3.8 of 100,000 interventions (33). Interestingly, this incidence was higher in women, and congestive heart failure and coagulopathy were the main factors associated with more severe lesions at the time of hospital discharge. However, previous publications provided an incidence of approximately 0.2% (34), which suggests that this figure may have been underestimated. This is either because patients with less severe neuropathies tend not to seek medical aid or because interventionists do not publish reports on it. On the other hand, the progressive development of the technique and the use of echography may have led to a drop in this incidence (35).

Among other diagnostic possibilities, RPH should be considered in a clinical picture compatible with LSP affection after a surgical procedure in an anticoagulated patient. Both spontaneous and postoperative RPH have
been described in anticoagulated patients, affecting the iliopsoas muscle and provoking lumbar and/or sacral nerve root compression. Its management is controversial because both surgical decompression and conservative treatment are viable options (36-40).

**CONCLUSION**

SCS may be a valid option for patients developing LSP secondary to ECMO application. Modern paresthesia-free SCS modalities such as burst stimulation may improve the relief provided by traditional stimulation in these patients.

**REFERENCES**


2. Garozzo D, Zollino G, Ferrari S. In lumbosacral plexus injuries can we identify indicators that predict spontaneous recovery or the need for surgical treatment? Results from a clinical study on 72 patients. *J Brachial Plex Peripher Nerve Inj* 2014; 9:1.


26. Roussel A, Al-Attar N, Alkhoder S, et al. Outcomes of percutaneous femoral cannulation for venoarterial extracorporeal mem-


