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CASE REPORT

Cervical dorsal rhizotomy for upper limbs spasticity. Case report

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Abstract
Spasticity is a prevalent symptom of upper motor neuron syndrome, becoming debilitating when hindering voluntary movement and motor function and causing contractures and pain. Functional neurosurgery plays a crucial role in treating severe spasticity. Despite extensive literature on SDR for lower limb spasticity, there is a scarcity of papers regarding the procedure in the cervical region to alleviate upper limb spasticity. This case report details a cervical dorsal rhizotomy (CDR) performed for upper limb spasticity, resulting in significant improvement in spasticity with sustained outcomes and low complication rates. Neuroablative procedures like CDR become an option to treat spasticity.

Keywords Spasticity · Rhizotomy · Upper limbs · Cervical · Case report

Introduction

Spasticity is a prevalent symptom of upper motor neuron syndrome, characterized by hypertonic muscles resisting passive motion in a velocity-dependent manner. This often coexists with hyperreflexia, clonus, Babinski sign, weakness, and incoordination, becoming debilitating when hindering voluntary movement, motor function, and causing contractures and pain. Typically, it follows insults to the central nervous system, arising from conditions like cerebral palsy, neurodegenerative disorders, brain, or spine trauma. The upper motor neuron pathophysiology involves a lesion in the first motor neuron, reducing inhibitory input to the second motor neuron, consequently increasing alfa motor neuron activity and exacerbating hypertonia and spasticity [6, 9, 11, 14, 16, 17].

Functional neurosurgery plays a crucial role in treating severe spasticity when conservative measures such as physical therapy, occupational therapy, anti-spasmodics, muscle relaxants, and botulinum toxin prove ineffective. Currently, there is consensus on selective dorsal rhizotomy (SDR) for ambulatory cerebral palsy (GMFCS II-III) and intrathecal baclofen pump (ITB) for non-ambulatory patients (GMFCS IV-V) [1, 3, 7, 13, 16, 17]. However, the high cost of ITB implantation and maintenance poses challenges, especially in low- and middle-income countries. Additionally, concerns arise if patients live far from specialized centers, compromising follow-up visits and pump maintenance. Recent reports suggest successful use of SDR for non-ambulatory cerebral palsy, indicating positive results in spasticity for both lower and upper limbs, along with some cognitive improvement. This improvement, noted above the level operated, is referred to as a suprasegmental effect [3, 13, 16, 17].

Despite extensive literature on SDR for lower limb spasticity, there is a scarcity of papers regarding the procedure in the cervical region to alleviate upper limb spasticity. This case report details a cervical dorsal rhizotomy (CDR) performed for upper limb spasticity.

Case report

A 7-year-old male child with cerebral palsy, history of premature birth, and a current weight of 16 kg presented with spastic quadriplegia (GMFCS V). The multidisciplinary team, including a neuropediatrician, neurosurgeon, and physiatrist, managed his care. Controlled epilepsy with oxcarbazepine (22.5 mg/kg/d BID) was noted.

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Fig. 1 Preoperative patient image revealing the severe spasticity, with pronounced elbow and fingers flexion

Conservative measures for spasticity included baclofen (2.5 mg/kg/d BID), botulinum toxin (last application 8 months ago), and ongoing physical and occupational therapy. A previous single-level laminectomy selective dorsal rhizotomy (SDR) in another hospital improved lower limb spasticity but left upper limb symptoms unchanged. Referred to our service, the child exhibited significant bilateral upper limb spasticity (modified Ashworth scale 4) and elbow and finger flexion, with contractures, impacting hygiene and care (Fig. 1).

Brain MRI revealed microcephaly, paquigyria (frontal and parietal lobes), gyral simplification, cortical thickening (more pronounced left hemisphere), cerebral hemisphere volumetric reduction with ventricular dilatation, and left thalamus volumetric reduction. Incomplete left hippocampus rotation was noted. Lumbar MRI showed no significant alterations post-laminectomy (Fig. 2).

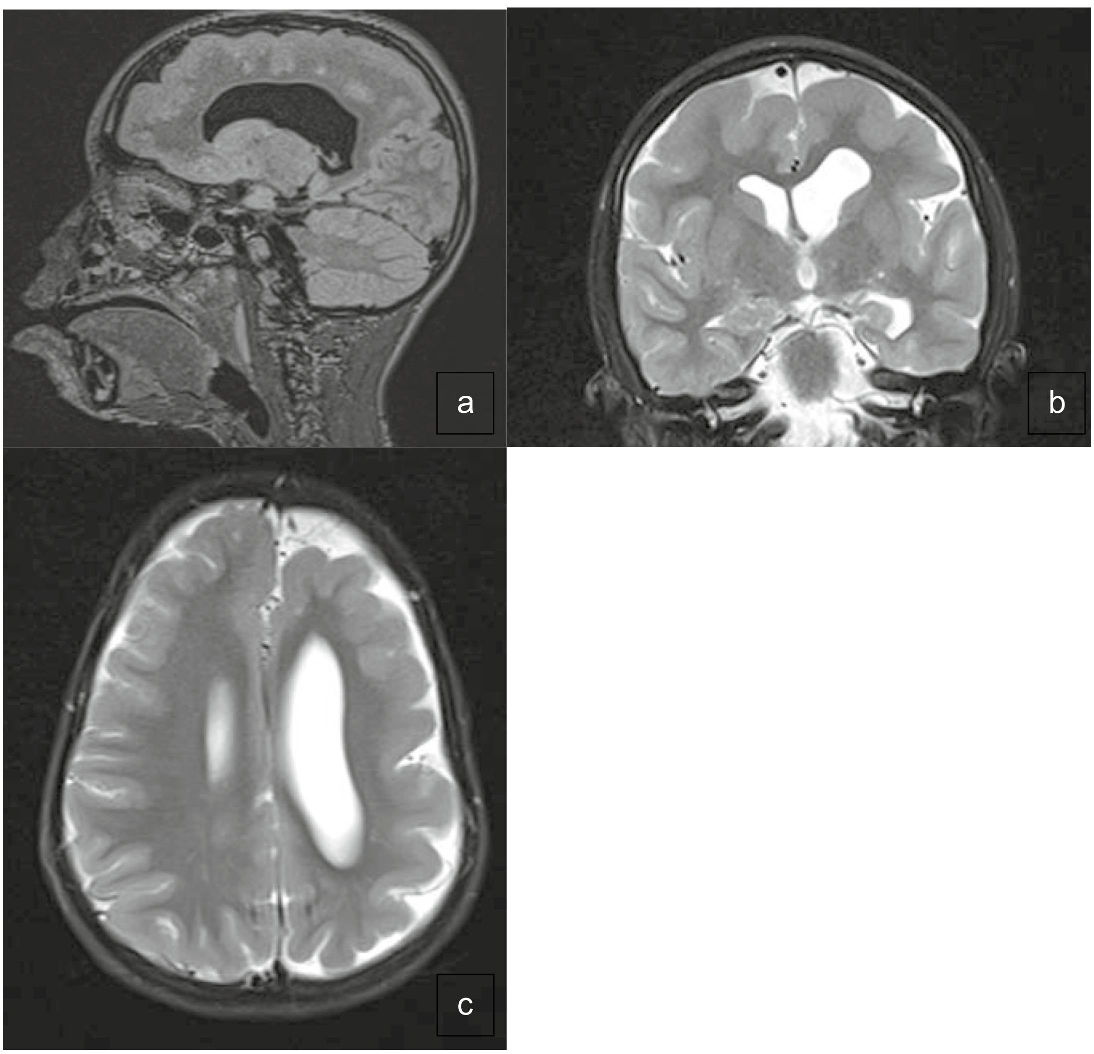

Fig. 2 Brain MRI showing the frontal and parietal lobes with paquigyria, gyral simplification, and cortical thickening (**a**–**c**). Incomplete left hippocampus rotation (**b**)

The intraoperative procedure (Fig. 3)

The patient, under general anesthesia and intraoperative electrophysiology, was positioned prone. A cervical midline incision and subperiosteal dissection were followed by C5, C6, and C7 laminotomy. The thecal sac was incised, and microscopy was introduced. Initially, C5, C6, and C7 roots were bilaterally identified through arachnoid dissection. The rootlets were then separated and stimulated with a bipolar probe, although electrophysiology did not definitively determine rootlet selection for rhizotomy. CDR sectioned 70% of the rootlets, with no motor evoked potential alterations observed throughout the procedure. Closure was performed layer by layer.

Post-operatively, spasticity decreased, particularly on the right side, enhancing elbow and finger extension. Discharge occurred on the fourth day without adverse events.

At the 6-month follow-up, the child showed significant spasticity improvement (modified Ashworth scale: left side 1, right side 2) with physical therapy and physiatric intervention. The GMFCS remained at level V (Fig. 4).

All healthcare providers and the mother expressed satisfaction with the procedure, noting improvement in patient hygiene and mobilization. They recommend such surgery for other patients with similar needs.

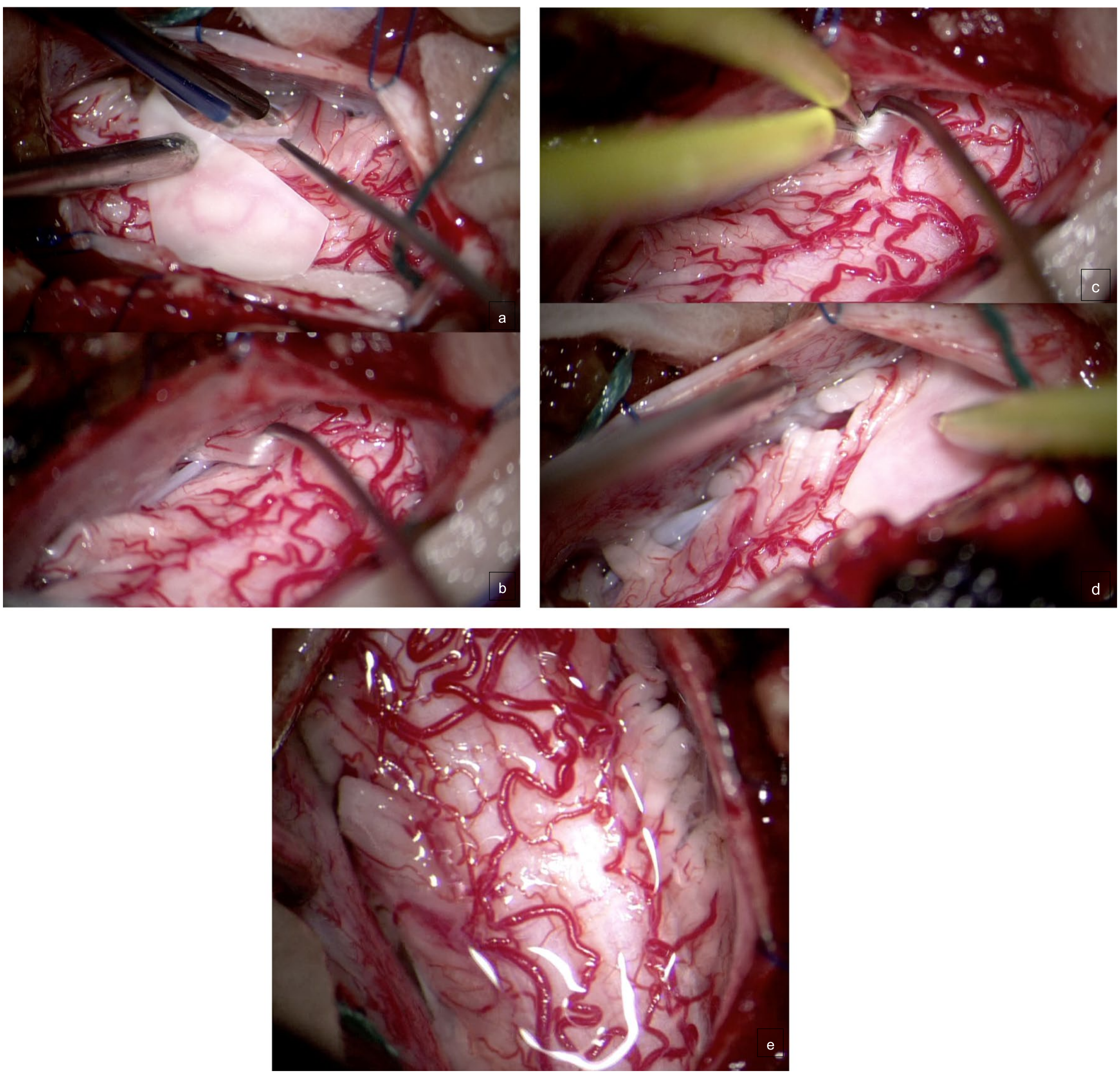

Fig. 3 Intraoperative images. The rootlets were then separated and stimulated with a bipolar probe (**a**, **b**). Coagulation and sectioning (**c**, **d**) of the rootlets. Cavity final aspect (**e**)

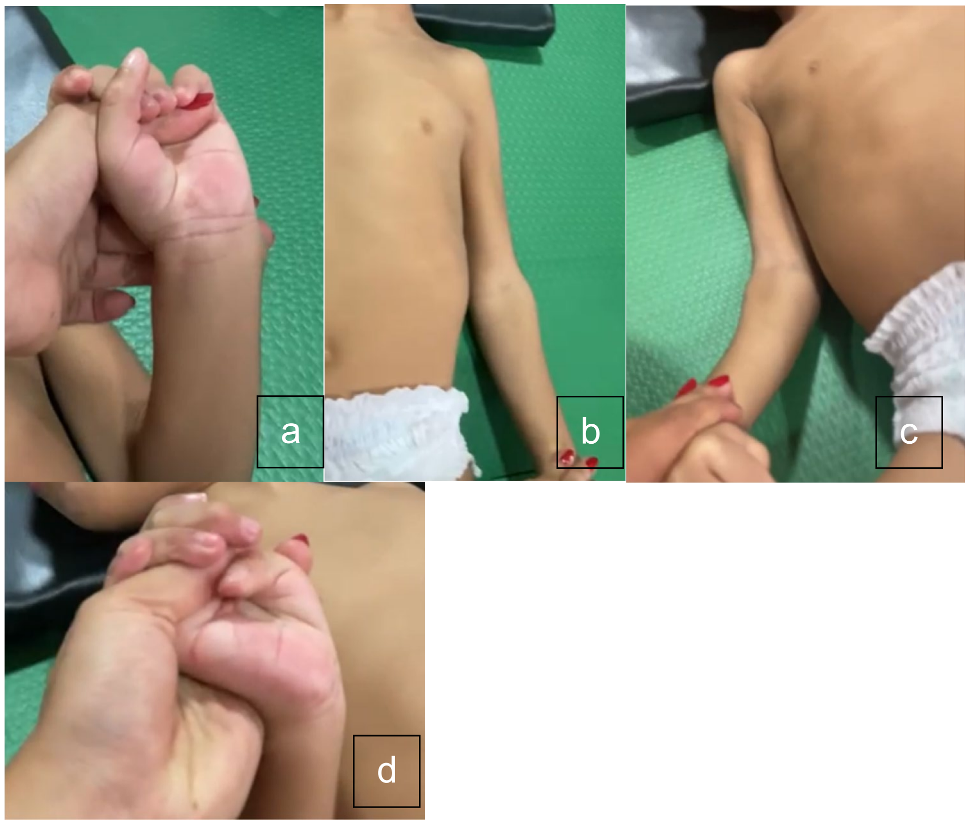

Fig. 4 Postoperative rehabilitation demonstrating the improvement in the spasticity in the elbow (**b**, **c**) and wrist (**a**, **d**)

Discussion

The management of spasticity depends on distinguishing between its general and focal manifestations and determining whether the treatment aims for reversible or permanent effects. Selective peripheral neurotomy (SPN) emerges as a viable option for addressing focal or residual spasticity affecting on or a few muscle groups. SPN entails a partial sectioning of motor nerve branches or fascicles, thereby disrupting the motor arc reflex and alleviating the coordinated action between agonist and antagonist muscles within the affected motor group. This intervention avoids involvement with sensitive fibers, thus mitigating the risk of neuropathic pain. Conversely, for addressing general spasticity, procedures such as rhizotomies, DREZL, and ITB therapy are typically favored [2, 5, 7, 8, 13, 15, 16].

Foerster initiated spasticity surgical treatment in 1908 with posterior rhizotomy, sectioning cauda equina's sensitive roots. However, severe sensitive disturbances, and eight deaths due to meningitis, led to its abandonment. Various surgical modalities followed, such as ventral rhizotomy, chordotomy, and myelotomy, all abandoned due to adverse effects. Dorsal rhizotomy resurged in the 1960s with Gros introducing SDR, and later, Ouaknine brought the microscopic technique to SDR. Fasano, in 1978, incorporated electrophysiology stimulation for root selection [1, 2, 8, 11, 12, 14, 17]. SDR targets Ia sensitive fibers, reducing input to alpha motor neurons and decreasing spasticity. Suprasegmental effects, observed in upper limbs, speech, and cognition after lumbar SDR, have been reported [8, 9, 11, 15–17].

Baclofen, acting as a GABAergic agonist, alleviates spasticity by enhancing inhibitory effects. However, oral baclofen faces challenges crossing the blood–brain barrier effectively. ITB addresses these limitations, reducing the required dose and mitigating side effects such as dizziness, memory disturbances, confusion, seizures, and respiratory depression. Despite its efficacy, ITB presents challenges due to device-related complications and high costs, thereby restricting its widespread use, particularly in low- and middle-income countries [3, 6, 7, 9, 17]. Alternative options for upper limb spasticity include dorsal root entry zone lesioning (DREZL), cervical dorsal rhizotomy (CDR), brachial plexus rhizotomy, and dorsal root entry zone irradiation [2, 5, 9, 10, 13–17].

DREZL interrupts myotatic reflex and nociceptive tract, mainly indicated for pain syndrome related to brachial plexus avulsion. Complications include neurologic deterioration and ataxia. CDR, similar to brachial plexus rhizotomy, has shown significant improvement with persistent results and fewer complications [3, 4, 8, 9, 12, 15–17].

Sindou et al. described 16 patients who underwent DREZL in the cervical cord due to pain and spasticity in the upper limbs. Fourteen out of 16 patients reported satisfaction with the procedure, but one patient experienced neurological deterioration, rendering them unable to stand or walk [14]. Hong et al. reported another series involving 9 patients who underwent DREZL, with 7 showing improvement in spasticity and 2 experiencing a worsening of symptoms. All patients who complained of pain achieved resolution of this symptom. One patient had a complication with transient ataxia [5]. Due to increased manipulation in the cervical cord, DREZL seems to present more complications than CDR.

Bertelli et al. reported on 13 patients who underwent brachial plexus rhizotomy (similar to CDR), showing significant improvement and persistent results in spasticity. They mentioned one case of recurrence due to incomplete resection of dorsal roots. No patient complained of motor deficit or ataxia. A suprasegmental effect was observed in two patients, resulting in improved speech. Seven patients showed better walking. No sensory disturbances or other complications were demonstrated, except for controlled cervical pain. These results were observed on the first postoperative day. In conclusion, the authors suggest C5 to C7 cutting for shoulder and elbow spasticity and include C8 if the hand presents symptoms. If only the hand and wrist are affected, C7 and C8 should be selected. This procedure should be preceded by orthopedic surgeries [2].

Guan reported successful C7 transfer with CDR to improve motor function and spasticity. Romanelli introduced dorsal root entry zone irradiation as a less invasive approach, requiring further studies for efficacy evaluation [3, 10, 12].

As a novel neuroablative therapy, CDR represents an additional technique for teams to consider, especially in low- and middle-income countries. CDR, with less manipulation and associated risks in the cervical cord, proves promising for upper limb spasticity, as shown in this report with good and persistent results and low complication rates. Future research will explore the efficacy of dorsal root entry zone irradiation as a less invasive method.

Author contribution All authors contributed to the study conception and design. Material preparation, data collection, and analysis were performed by Ítalo Teles de Oliveira Filho, Paulo Cesar Romero, and Alexandre Pingarilho Rezende. The first draft of the manuscript was written by Ítalo Teles de Oliveira Filho and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Declarations

Ethics approval Ethical approval was waived by the local Ethics Committee in view of the retrospective nature of the study, and all the procedures being performed were part of the routine care.

Informed consent Written informed consent was obtained from the parents.

Consent for publication The parents have consented to the submission of the case report to the journal.

Competing interests The authors declare no competing interests.

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