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Intracerebroventricular Baclofen Therapy for Chronic Pelvic Pain in Cerebral Palsy: A Case Report

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Introduction: Although pain is frequent among patients with cerebral palsy (CP), vulvodynia is an uncommon finding. Its diagnosis is underrated in this population and its treatment is challenging, due to uncertainty in pathophysiological mechanisms of the symptom. The management of this rare combination of relevant sensorial and motor components requires a skilled multidisciplinary team capable of offering and testing all available forms of treatment, since this pain carries high levels of suffering and incapacity, and patients with CP usually have many other chronic and severe comorbidities.

Case presentation: This is a 14-year-old girl with mixed (hyperkinetic and spastic) CP, with minimal cognitive impairment, and classified as Gross Motor Function Classification System (GMFCS) level V. She failed all available conservative treatments for a longstanding and disabling vulvodynia. Local injections, including toxin botulinum failed to provide relief. After responding to an intrathecal baclofen trial, a baclofen pump was implanted with the catheter positioned within the third ventricle through navigation guidance. She experienced complete pain relief, and spasticity improvement as well. During the 6 months of follow-up, patient was pain free. Her quality of life also improved with the therapy during the follow-up period.

Conclusion: Intracerebroventricular baclofen therapy may be a viable option for managing chronic pain associated with spastic-dystonic symptoms in cerebral palsy.

Keywords: cerebral palsy; vulvodynia; intraventricular baclofen; dystonia; spasticity

INTRODUCTION

Pain is a common finding in cerebral palsy (CP) patients, especially among females, older children, non-ambulants and in children with predominant hyperkinetic (dystonic or choreiform) manifestations [1]. Its prevalence ranges from 14% to 76%. A prospective cross-sectional study showed a 79% prevalence of acute pain and 76% of chronic pain in a mixed (hyperkinetic and spastic) CP population, and identified contributors to central sensitization and pain chronification in these patients such as: high frequency and extensive distribution of hyperkinetic movements; presence of hip dislocations, scoliosis, fractures and deformities; history of multiple invasive procedures (injections, bone and soft tissues surgeries); cumulative lesions during physical therapy (fractures, subluxations, stretching, sprains); bad positioning, transferring or walking; constant pressure on regions under maladapted wheelchairs, orthotics and prosthetics [1,2]. The higher the Gross Motor Function Classification System (GMFCS) grade, the higher the prevalence of pain and its multiple location [1].

The most frequent reported locations of pain are the lower limbs, and the thigh/ groin/ pelvic region is referred by 35 % of the individuals [1]. Vulvar pain global prevalence is estimated in 3 % to 15% of the general population, with no data specific for CP patients.

The diagnosis of vulvar pain is often overlooked in the general population, possibly due to sociocultural and religious factors, lack of proactive screening by caregivers, or limited awareness among healthcare providers. When this scenario is transposed to the population with CP, with multiple health issues, attention to this complaint can be diluted. A subgroup of vulvar pain – vulvodynia - requires more efforts to be identified, since it is an exclusion diagnosis. To meet the current diagnostic criteria for vulvodynia, the vulvar pain must be present or recur for more than 3 months, without any identifiable etiology (idiopathic) and with a few potential associated factors. Among them are some comorbidities and other pain syndromes (fibromyalgia, painful bladder syndrome, irritable bowel syndrome), genetics, hormonal factors, inflammation,

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musculoskeletal (pelvic muscle overactivity, myofascial), neurologic mechanisms (central and peripheral), psychosocial factors and structural pelvic defects. The intensity of pain is usually high and the impact over life quality may be catastrophic. To the present, its treatment forms have inconsistent results, and an individualized therapy is usually required [3].

In this manuscript, we report an uncommon case of an isolated vulvodynia in a 14yo CP patient, successfully controlled using an uncommon approach: intracerebroventricular baclofen (IVB) therapy.

CASE REPORT

A 14yo female patient was diagnosed with mixed (hyperkinetic and spastic) CP, with minimal cognitive impairment and classified as GMFCS 5. She presented a primary, chronic, persistent, spontaneous and localized vulvodynia that became severe in the last 2 years, with basaline pain intensity scored in Visual Analogue Scale (VAS) 9 out of 10. The pain was intensified during miction (VAS 10/10) leading her to avoid drinking water. No kidney disfunction was noted during the initial evaluation. She presented a dystonic-spastic state with Modified Ashworth Scale (MAS) of 3 on in right upper extremity (RUE), MAS 2 in left upper extremity (LUE) and 4 in both lower extremities (BLE). Bilateral ankle clonus was noticed. Her vulvar pain was not relieved by oral analgesics (dipyrone, acetaminophen, non-steroids anti-inflammatories, opioids), urinary tract analgesic (phenazopyridine hydrochloride), tricyclics antidepressants (nortriptyline), anticonvulsants (gabapentin, pregabalin) and oral muscles relaxants (baclofen and triexiphenidine). Topical treatment with lidocaine had no response. Regional analgesic blocks made no improvements and botulinum toxin type A applications paradoxically got the pain worsened (probably because of central sensitization). Pelvic physiotherapy and psychological treatment also did not have any results on the pain control. Sessions of transcranial magnetic stimulation were also carried out, with temporary and minimal improvements in pain scores. After exhausting the available treatment options at that time, it was considered that a possible mechanism for the pain could be dystonia of pelvic muscles associated to central sensitization pain. Then an intrathecal baclofen trial was proposed.

Patient was evaluated using VAS, McGill Pain Questionnaire, Douleur Neuropathique 4 Questionnaire (DN4) for pain, MAS for spasticity and World Health Organization Quality of Life Assessment - short form (WHOQOL-Bref) for quality of life. Just before the procedure, patient scored 23/78 in McGill Pain Questionnaire, 10/10 in VAS and 6/10 in DN4. In McGill's sensorial-discriminative category, the patient reported 13 points, with the following pain descriptors: pulsing and throbbing; jumping; pricking; cutting; pressing; tugging; hot and burning; tingling, smarting and stinging; sore; tender. The affective-motivational category summed 5 points, and pain was described as exhausting; sickening; frightful; vicious; blinding. For the cognitive category, she scored 1 point (unbearable pain). The score in miscellaneous category was 4 points: penetrating; tight and tearing; excruciating. All these features delineated a neuropathic quality for the pain and mechanisms of central sensitization.

A single-shot trial with barbotage infusion of 50mcg of a compound intrathecal baclofen was performed in lumbar spine (Baclofeno Intratecal 50mcg/mL - Cytopharma™ – Belo Horizonte - Brazil), with the patient in lateral decubitus and under conscient sedation because of the involuntary movements. It was observed a complete resolution of the pain for 48h (VAS 0 out of 10). Spasticity improvement was also noticed. After that period, she returned to her baseline pain and spasticity. After this successful trial with 50 mcg of intrathecal baclofen, permanent therapy was indicated using an intracerebroventricular approach [4,5]. The approach was considered because of an already present moderate scoliosis that might require future correction and would displace the spinal catheter. Most importantly, the intracerebroventricular delivery of baclofen has a reported potential better effect compared to the spinal intrathecal route [4]. The procedure was performed under general anesthesia and followed similar technical steps to a ventriculoperitoneal (VP) shunt. The pump catheter (Medtronic Ascenda™) was connected to a standard VP shunt intraventricular catheter from HPBio™ – São Paulo - Brazil) placed into the third ventricle (Figure 1). The procedure was guided by neuronavigation (BrainLab™ Neuronavigation System). The distal Ascenda™ catheter was then tunneled underneath the subcutaneous layer and connected to a Medtronic Synchromed II™ pump with 20mL reservoir (Synchromed II pump – Medtronic ™- Minneapolis, Minnesota, USA) which was positioned on the right lower abdominal wall.

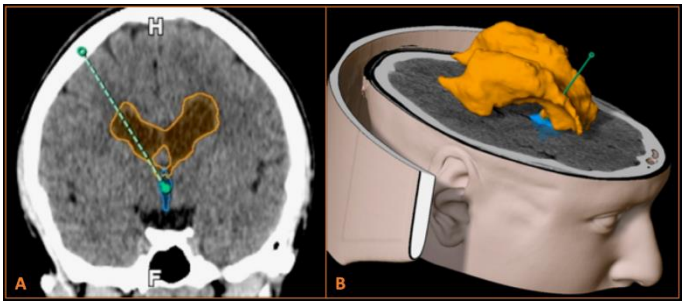

Figure 1- Neuronavigation planning for proximal catheter tip positioning into the third ventricle.

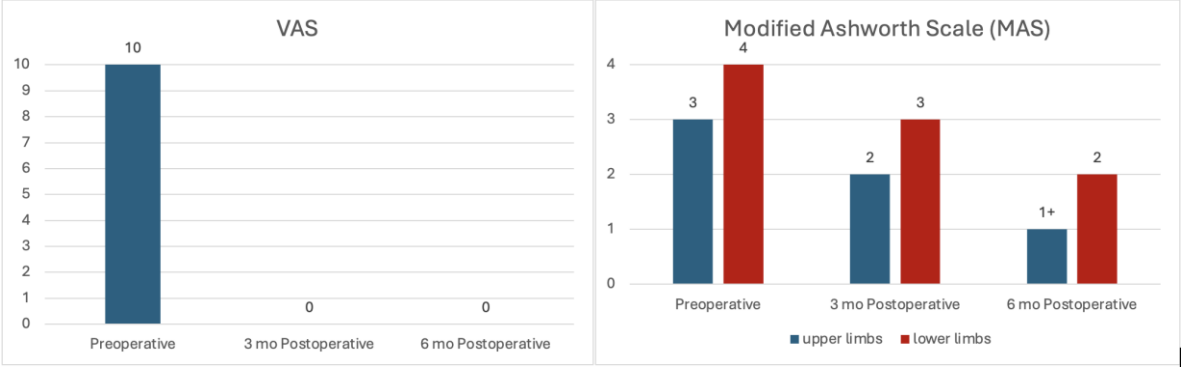

Figure 2- Evolution of VAS rating and MAS grading

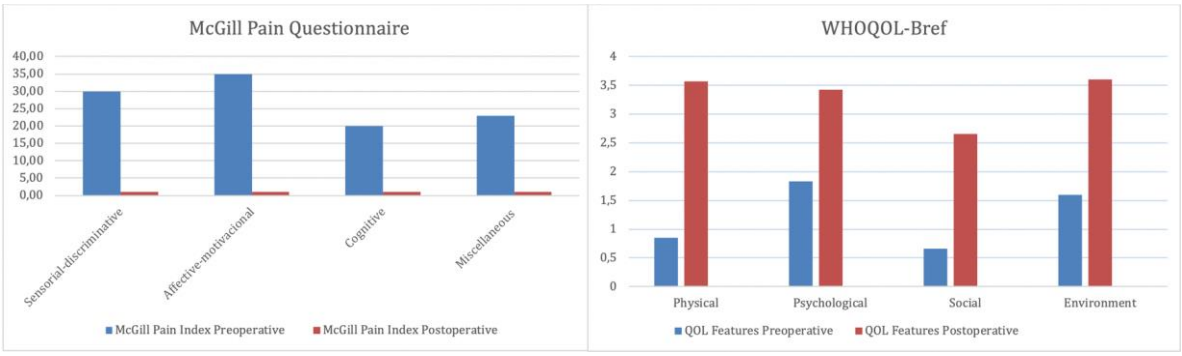

Figure 3 - Evolution of McGill Pain Questionnaire and WHOQOL-Bref scores before and 6 months after the procedure.

Even with a very small initial baclofen dose (35mcg/24h), a complete vulvodynia improvement was achieved (VAS 0/10) along the first postoperative 4 hours, coinciding with the priming pump bolus duration. Intracerebroventricular baclofen initial infusion dose was set to 1mcg/Kg (6) in simple continuous mode. Through the first week, patient reported anorexia and nausea. The pump infusion was then reduced to 25mcg/24h, a gastric protector was administered, and all symptoms were resolved. She didn't experience any pain with the drug titration. Intending to improve also the spasticity, weekly infusion increments were restarted, with slowly increasing dosage. After the complete and persistent vulvar pain resolution with very low baclofen doses, finally the dystonic-spasticity improvement was observed with higher doses (110mcg/day, in simple continuous infusion mode). In the 6 months follow-up, evaluation with MAS for RUE remained in 3 points, while 1+ point for the LUE and 2 for BLE (Figure 2). Functional aspects and quality of life, features of chronic pain as valuated respectively by WHOQOL-Bref and McGill Pain Questionnaire also presented expressive improvement (Figure 3).

DISCUSSION

This CP patient met the diagnostic criteria for vulvodynia associated with motor dysfunction of pelvic floor after extensive etiologic investigation, with signs of central sensitization pain mechanisms that justified its high intensity, chronicity, distress and refractoriness to the usual treatment. Her neurological findings of severe spasticity and dystonia, mobility and deformity issues pointed to a motor dysfunction of pelvic floor that promoted and amplified a vulvar pain. In a bidirectional manner, the pain exacerbated the involuntary movements, keeping this vicious cycle [3].

In our report, very low doses of baclofen delivered by the intracerebroventricular route promoted sustained complete pain relief, even before any motor improvement. Analgesia promoted by intraventricular baclofen has not only a spinal component but also an action in structures such periaqueductal gray matter (PAG) [7,8]. Microinjections of baclofen in the caudal PAG, in the brainstem at sites lateral to the midline or near the raphe magnus promotes analgesic effects in mouse [9]. This dual mechanism of pain relief (spinal and supraspinal) supported our choice of an intraventricular approach in cases where both local antinociceptive action in brainstem and motor relaxation

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were desired. Some cases of lack of response in intrathecal baclofen (ITB) infusion showed improvement with intraventricular baclofen, suggesting that this route can be more effective specially for dystonic patients [4]. The catheter tip should be placed into the third ventricle [6,10], where infused baclofen directs down the aqueduct and fourth ventricle, passing through the PAG, then enters the subarachnoid space and distributes over the cerebral convexities, where baclofen may produce its effect in treating dystonia [11]. In an experimental study, beagles showed toxic reactions related to doses superior to 250mcg/day, death with 1000mcg/day, with no ependymal damage [12]. Doses of 135mcg/day or less showed to be secure [12]. In humans Albright reported IVB safe doses of 2012mcg/day without major complications [4]. The most common complications related to IVB were infection, catheter misplacement/migration and pump malfunction/pocket issues [13]. In his study, 10 out of 20 patients (50%) with IVB presented just one complication. One death was reported related to respiratory failure, with 37% presenting complications related to the procedure, compared to 48% of ITB [5]. CSF leak occurred in ITB patients in 21% and in IVB ones in 6,7%. Barry-Albright dystonia scales scores decreased in patients with ITB in 7 points compared to 9 points in patients with IVB [5].

Intracerebroventricular baclofen therapy seems to not just improve patient quality of life (8), but also, their caregivers, as reported in other spasticity methods, like selective dorsal rhizotomy [14].

Just after the IVB therapy, the reported patient experienced anorexia. Controversially, an animal study showed increased food intake after IVB [15]. Another interesting point learned from our patient is that the pain improvement preceded motor improvement, supporting a supraspinal mechanism of action. This observation is consistent with animal studies demonstrating analgesic effects of baclofen in periaqueductal regions [9]. With increased doses, finally MAS improved was noticed, but, first in the lower extremities and not in the uppers, as expected from a more cranial infusion. The improvement of the upper extremities came after the lowers, and in a smaller proportion. The dynamics of the CSF circulation can probably justify why this sequence was observed.

CONCLUSION

Intracerebroventricular baclofen therapy may offer effective pain relief in patients with cerebral palsy and chronic pelvic pain. Pain control can be achieved at low doses, but higher doses may be necessary to address hypertonia, especially in the upper limbs.

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DISCLOSURES

Ethical approval

This study was conducted in accordance with the principles outlined in the Declaration of Helsinki. Institutional approval was not required as per the guidelines of the Declaration

Consent to participate

The patient's legal guardians provided consent for the use of his information and images in this publication

Conflict of interest

Bernardo Assumpcao de Monaco and Eduardo Joaquim Lopes Alho are speakers/ proctors for Medtronic – Brazil. All the other authors declare no direct conflict of interests.

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Artificial intelligence

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CONTRIBUTIONS

-**Guilherme Corsaletti Gregório**: Conceptualization, Data curation, Formal Analysis, Investigation, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing
-**Fabio Okuda Furokawa**: Investigation, Resources, Supervision, Writing – original draft, Writing – review & editing
-**Eduardo Joaquim Lopes Alho**: Supervision, Visualization, Writing – review & editing
-Luciano Furlanetti: Investigation, Validation, Visualization, Writing – review & editing
-Bernardo Assumpção de Monaco: Supervision, Validation, Visualization, Writing – review & editing

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