

*CASE REPORT*

## **Rehabilitation Management of a Child with Severe Pain after Bacterial Meningoencephalitis: A Case Report**

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### **ABSTRACT**

**Introduction:** Pain is common in children with meningoencephalitis and can cause negative consequences. Therefore, this study aimed to report the case of a child who experienced severe pain after *Acinetobacter* meningoencephalitis infection and was subjected to rehabilitation management.

**Case Presentation:** A 1-year-4-month-old child was brought to the rehabilitation outpatient clinic with stiffness and severe pain after *Acinetobacter* meningoencephalitis. The symptoms observed include post-repair amputated tongue, hypertonus muscles, spasticity, and asymmetrical posture. After rehabilitation program, pain significantly reduced to moderate intensity.

**Case discussion:** Pain can cause a vicious cycle of spasms or spasticity that further worsens the condition pain. Multiple organ system impairments may contribute to overall pain experienced by the patient. Therefore, a multidisciplinary team is needed to evaluate and manage the multiple sources of pain. Multimodal rehabilitation interventions consisting of 24-hour postural management, spasticity medication, ultrasound therapy, massage, stretching, and orthosis prescription may help to manage pain.

**Conclusion:** Pain after meningoencephalitis may be caused by multiple sources and requires multidisciplinary and multimodality approaches for effective management.

## ABSTRAK

**Latar Belakang:** Nyeri sering ditemukan pada anak dengan meningoensefalitis dan dapat menimbulkan berbagai dampak negatif. Laporan kasus ini menjelaskan mengenai seorang anak dengan nyeri intensitas berat setelah meningoensefalitis *Acinetobacter* yang mendapatkan program rehabilitasi.

**Deskripsi kasus:** Seorang anak usia 1 tahun 4 bulan dibawa ke poliklinik rehabilitasi dengan keluhan kaku dan nyeri berat pasca *Acinetobacter* meningoencephalitis. Ia memiliki riwayat amputasi lidah pasca bedah, otot hipertonus, spastisitas, dan postur asimetris. Setelah program rehabilitasi, nyeri berkurang menjadi intensitas sedang.

**Diskusi kasus:** Nyeri dapat menyebabkan lingkaran setan antara spasme otot-spastisitas akibat nyeri yang selanjutnya memperberat nyeri, dan seterusnya. Gangguan berbagai sistem organ dapat menjadi sumber nyeri. Tim multidisiplin diperlukan untuk mengevaluasi dan mengelola sumber nyeri multipel. Intervensi rehabilitasi dengan modalitas yang terdiri dari manajemen postural 24 jam, medikamentosa, terapi *ultrasound*, pijat, peregangan, dan peresepan ortosis dapat membantu manajemen nyeri.

**Kesimpulan:** Nyeri pasca meningoensefalitis dapat disebabkan oleh berbagai sumber nyeri yang ditangani oleh pendekatan multidisiplin dan multimodalitas.

**Kata kunci:** nyeri, meningoensefalitis, rehabilitasi

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## INTRODUCTION

Meningoencephalitis is an inflammatory disease of the brain membrane and parenchyma due to infections by viruses, bacteria, fungi, and parasites.<sup>1,2</sup> Pediatric *Acinetobacter* meningoencephalitis is relatively rare.<sup>11</sup> Among all cases, the majority are often subjected to

neurosurgical procedures for the removal of space-occupying lesions from the brain parenchyma.<sup>3,4</sup> Despite adequate treatment, bacterial meningoencephalitis in children still has a high risk, leading to neurological deficits sequelae with up to 27% prevalence.<sup>5</sup> The most commonly reported sequelae are developmental delay<sup>1,5</sup> and a significant number of children affected by meningitis often have one or more permanent disabilities.<sup>6</sup> Additionally, meningitis ranked sixth in the top causes of disability-adjusted life years (DALYs) among children under 10 years of age in 2019.<sup>7</sup>

Pain is a common symptom in children with brain lesions, attributable to multiple sources and often undertreated.<sup>8,9,10,11</sup> Effective management is crucial in these patients as pain can cause numerous negative consequences and hinder

the effectiveness of rehabilitation program.<sup>8,12</sup> There are limited studies reporting about management of pain after meningoencephalitis. Therefore, rehabilitation management for children who survived after meningoencephalitis is challenging.<sup>6</sup> This study reported the case of a child who experienced severe pain after *Acinetobacter* meningoencephalitis and was subjected to rehabilitation management in the outpatient setting.

## CASE DESCRIPTION

A 1-year-4-month-old child was admitted to the hospital following repeated seizures. After the seizures, there was a decrease in consciousness and stiffness of the trunk and extremities. The right knee was fixed in extension and the right ankle was fixed in plantarflexion due to severe spasticity. Avulsion tongue injury also occurred during seizures and was managed by operative repair. Subsequently, a brain CT scan showed cerebral atrophy with ventricle dilatation and hypodense area near the anterior and posterior horns of both sides (figure 1). The cerebrospinal liquor analysis suggested *Acinetobacter* sp. infection. A pediatric neurologist diagnosed the condition to be *Acinetobacter* Meningoencephalitis, leading to hospitalization for two months. During the inpatient period, the patient was also referred to the Physical Medicine and Rehabilitation Specialist. Botulinum toxin injection was given, followed by stretching and serial splinting of the quadriceps and gastrocnemius muscles.

A week after discharge from the hospital, the patient was brought by the parents to the Rehabilitation Outpatient Clinic. This time, the parent's chief complaint was stiffness of

the trunk and extremities. The seizure was controlled by anti-convulsant medication including baclofen 3x2.5 mg, valproic acid 2x75 mg, and phenytoin 2x35 mg. Before seizures, developmental milestones were typical, and after meningoencephalitis, gross, fine, and oral-motor, as well as communication skills were lost. The patient was unable to lift the head or roll over, the fingers were in a flexed position and could not be used to reach objects. The previous spoken words had been lost and could only cry in a small voice. Regarding the oral-motor skill, the patient could not drink and eat through the mouth efficiently. There were also periods of choking, hence, the nasogastric tube was maintained.

Physical examination indicated inadequate environmental awareness with no eye contact and sub-optimal auditory response towards the source of sound. The patient constantly appeared to be in pain. Faces, Legs, Activity, Cry, Consolability (FLACC) Pain Scale was 9, indicating severe pain, discomfort, or both. The posture appeared asymmetrical with a flexed trunk to the right side, flexed elbows, and extended legs (figure 1). Furthermore, poor head, neck, and trunk control was observed. When held in a sitting position by elbow approximation, the left shoulder was lower than the right side, the thoracic spine was convex to the right side, and the left pelvis was lower than the right side.

Due to the severely painful appearance, all the factors that could contribute to the sources of chronic pain were examined, including spasticity, high muscle tones, joint stiffness, malalignment, and post-surgical pain. On the musculoskeletal examination of the trunk, high postural tones of the extensor muscles were observed. Palpation of the trunk indicated tightness of the erector spinae,

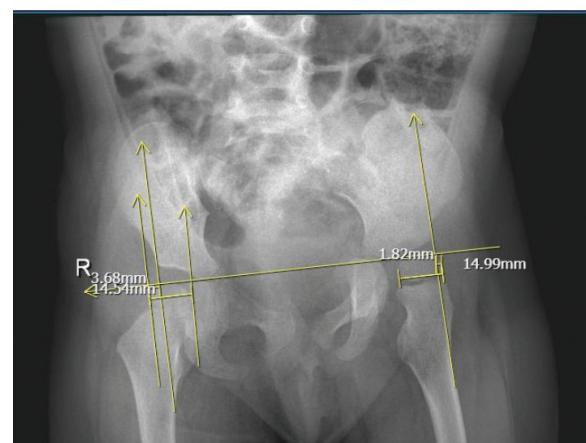
latissimus dorsi, and quadratus lumborum. The upper extremities tended to stiffen with shoulder, elbow, wrist, and phalanges in a flexed position. There was spasticity in both shoulder adductor and elbow flexor with Modified Ashworth Scale (MAS) of 2. The lower extremities tended to stiffen in the hip and knee in an extended position as well as the ankle in a plantar flexed position. There was spasticity with MAS of 2 in the hip and knee extensor muscles.

Intraoral examination showed a partially amputated tongue, mouth-breathing, drooling, pooling of saliva, stiffened temporomandibular joint, and inadequate tongue-jaw-neck movement. Respiro-motor examination showed symmetrical chest wall movement, no contraction of accessory muscle, no retraction of the chest wall, normal breathing sound, and no phlegm in auscultation.

The radiologic examination of the thoracal spine showed normal arrangement and position. Furthermore, the pelvic x-ray demonstrated erosion of the right femoral head, femoral column, and ischium, with widening of the right hip joint space and hyperdense of the soft tissue surrounding the right hip joint. The Reimer migration index of the right and left hip were 25% and 13%, respectively (figure 2), suggesting right hip septic arthritis based on the radiologist's assessment.



**Figure 1. CT-scan of The Brain**



**Figure 2. Radiograph of Pelvis.**

According to the examination results, rehabilitation program was prioritized to reduce pain intensity. Multidisciplinary collaboration is needed in pain management, especially in the diagnosis of pain sources. The patient was referred to the orthopedic surgeon for evaluation of possible hip septic arthritis. Evaluation by hematology infective markers did not support the diagnosis, hence, the surgeon only performed observation without any further debridement plan. An otorhinolaryngologist was further consulted for flexible endoscopic evaluation of swallowing

(FEES) to detect any risk of aspiration. The patient was only diagnosed with oral dysphagia without impaired pharyngeal swallowing or aspiration.

To manage muscle spasms and reduce muscle tightness, ultrasound therapy, followed by gentle massage and stretching for trunk flexor, shoulder adductor, elbow flexor, and knee extensor muscles was performed two times a week. Ultrasound therapy was given in continuous mode, frequency of 3 MHz, an intensity of 1-1,2 W/m<sup>2</sup>, and 5-10-minute durations for each region. Stretching was given in 10 repetitions and 10 seconds of hold for each muscle. Two types of orthoses were prescribed to support the proper alignment and prevent contracture. A resting hand splint was used on the left hand and solid ankle foot orthoses were used on both ankles.

The mother was educated on how to perform 24-hour postural management at home, which consisted of massage, stretching, proper positioning, and the use of assistive device. Other activities include applying a warm pack, gentle massage, and stretching of the tightened muscles every day, with proper prone, sitting, and sleeping positions. A wedge pillow was used to support the prone position. The patient was seated on the mother's lap while manually holding hand in front of the body in a sitting position (figure 3). The sleeping position was preferably on the left side until independent rollover could be achieved in the most comfortable position. When transferring or carrying the patient, the mother was asked to adopt an extended position, with the chin poked forward and arms turned in at the shoulders and flexed against the body. One hand may be fisted, while the hips and legs are turned in and partially extended.



**Figure 3. Child's Posture Before (Left) and After Rehabilitation Program (Right); The Sitting Position on Home Program (Middle).**

After four weeks of rehabilitation program, some improvements were observed. The FLACC Pain Scale decreased from 9 to 5 indicating moderate pain, discomfort, or both. The trunk posture appeared more symmetrical (figure 3), the elbows were flexed, and the legs remained extended, but easier to be positioned. Head and neck control

was achieved for several seconds when held in a sitting position by elbow approximation. Furthermore, the shoulder and pelvis appeared more symmetrical and the thoracic spine had better arrangement. The patient still had no eye contact but started to turn the head towards the mother's soothing voice.

**Table 1. Faces, Legs, Activity, Cry, Consolability (FLACC) Pain Scale**

FLACC Pain Scale Category	Before rehabilitation program	After rehabilitation program
<b>Face</b>	2 – Frequent to constant frown, clenched jaw, quivering chin	1 – Occasional grimace or frown, withdrawn, disinterested
<b>Leg</b>	2 – Kicking or legs drawn up	1 – Uneasy, restless, tense
<b>Activity</b>	2 – Arched, rigid, or jerking	1 – Squirming, shifting back/forth, tense
<b>Cry</b>	2 – Crying steadily, screams or sobs, frequent complaints	1 – Moans or whimpers, occasional complaint
<b>Consolability</b>	1 – Reassured by occasional touching, hugging, or “talking to”, distractible	1 – Reassured by occasional touching, hugging, or “talking to”, distractible
<b>Total score</b>	9	5

## DISCUSSION

This study presented the case of a 1-year-4-month-old child in the Outpatient Rehabilitation setting who had severe stiffness of the body and trunk. The patient had just been discharged from the hospital one week before with a diagnosis of *Acinetobacter* Meningoencephalitis. Subsequently, all developmental milestones were regressed.

The recovery after meningoencephalitis varies from complete to incomplete. A systematic review suggested that rehabilitation interventions among pediatrics and adults with infectious encephalitis are beneficial.<sup>1</sup> Rehabilitation intervention during the acute period after encephalitis needs to provide a safe

and optimal environment to facilitate the process of recovery and developmental stimulation.<sup>1,13</sup> Despite developmental regression, pain in the patient was prominent and important to address. In this case, rehabilitation intervention was prioritized for pain management before the developmental stimulation.

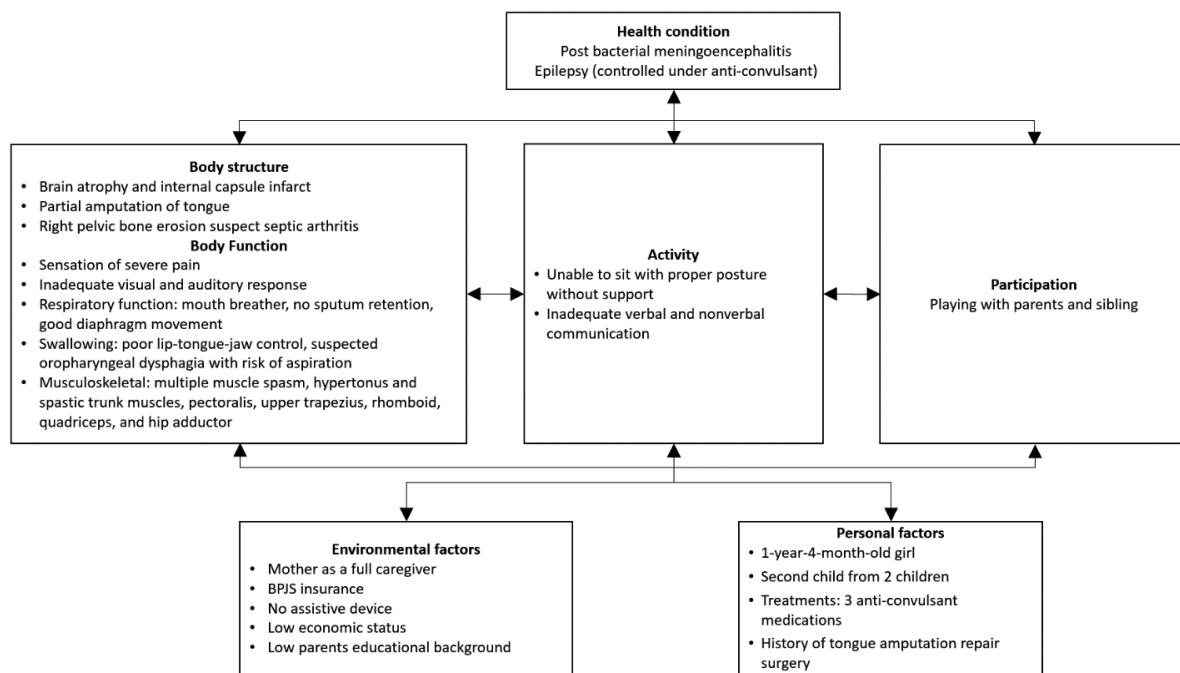
Pain is a significant problem frequently observed in children with brain lesions but is often unrecognized and undertreated. Difficulties in identifying pain might be due to the potential origin from multiple organ systems, and children may have communication problems.<sup>8-11,14</sup> However, the need for immediate and appropriate pain-relieving treatment remains crucial, as children may have significant negative experiences.<sup>11</sup> Pain

potentially causes a vicious cycle of spasms or spasticity that further worsens the condition.<sup>8</sup>

Face, Legs, Activity, Cry, and Consolability (FLACC) scale was used to quantitatively measure the pain intensity since the child was unable to communicate verbally. This scale defines five pain behaviors in young children who cannot self-report or verbalize the presence or severity.<sup>11</sup> A study reported high interrater reliability and validity of FLACC scale in young children aged 1-4 years with cerebral palsy during early developmental intervention programs.<sup>15</sup>

Pain in children with brain lesions can vary in duration from acute to chronic.<sup>8,11</sup> It may also be classified as primary or secondary. Multiple organ system impairment can be the source

of secondary pain, including musculoskeletal, visceral, peripheral nervous system, or central nervous system.<sup>8</sup> In this case, factors that could contribute to pain problems were assessed according to the International Classification of Functioning, Disability, and Health (ICF) (figure 4). Relevant to the biopsychosocial model, ICF offers a framework for describing and organizing information about health-related components and contextual factors in facilitating and/or hindering functioning. In this context, functioning is defined as “what a child with a health condition can do every day”.<sup>10</sup> Based on the ICF model, the impairments of body structures and functions that may cause pain in the patient include erosion on the right pelvis bone, hypertonus and spastic of multiple muscles, as well as multiple muscle spasms.



**Figure 4. ICF Describing Functional Components of The Patient.**

Pain in children with brain lesions is often attributed to the chronic secondary type from musculoskeletal origins,<sup>8,16</sup> while the primary source such as complex regional pain syndrome remains rare.<sup>8</sup> The musculoskeletal sources of pain include deformity, increased muscle tone and spasticity, hip dislocations, or scoliosis.<sup>8,16</sup>

Spasms can be a direct source of recurring pain through muscle contractions leading to vascular compression, which causes muscles to consume large amounts of oxygen while working under ischemic conditions, eventually activating the nociceptors.<sup>8</sup> Pain may increase in muscle spasticity, creating a vicious cycle. Spasticity, muscle weakness, and soft tissue changes in spastic children may lead to joint deformities and misalignment, especially in weight-bearing joints.<sup>17</sup> The joint misalignment and inability to change position increase the risk of having postural asymmetries in both sitting and lying positions, increasing the risk for pain.<sup>18</sup> This may contribute to musculoskeletal complications and secondary pain sources, such as hip dislocation, scoliosis, and joint contractures.<sup>17</sup> Postural asymmetries can also cause pain due to the impingement of ribs against the elevated side of pelvis.<sup>18</sup>

The target of rehabilitation program is to maintain proper positions in sitting, supine, or prone, that can prevent further complications. To achieve these proper positions, pain sources were managed in the patient. Reducing pain can make it easier for the caregiver to position the child in a comfortable way and for a long duration, further facilitating developmental stimulation. Therefore, the most important part of rehabilitation program was 24-hour postural management at home, consisting of stretching, positioning, and the use of an assistive device. To facilitate home-based

postural management, the child also received oral medication, ultrasound therapy, massage, stretching, and orthosis prescription to help reduce spasticity and muscle spasms. Botulinum toxin injection and serial casting were also given in the inpatient setting.

Options for spasticity-related pain include botulinum toxin injection or oral medication.<sup>6</sup> Botulinum toxin injection selectively inhibits the release of acetylcholine from peripheral nerve terminals at neuromuscular junctions. The effect generally lasts for 3-5 months, as the nerve terminal develops new synaptic terminals.<sup>16,19</sup> The patient in this case received a botulinum toxin injection while being hospitalized. After the muscle spasticity had been released, the joint arrangements were improved and maintained by serial splinting, which provided a prolonged stretch, offering biomechanical benefits and inhibiting spasticity.<sup>20</sup> The ankles' range of motions became plantigrade, and serial splinting was replaced by the orthosis. The patient also continued the anticonvulsant medication including baclofen. Aside from the anticonvulsant effect, baclofen is one of the oral medications recommended for muscle relaxation that can be used for long-term pain relief.<sup>16,19</sup> This combination of treatment, comprising botulinum toxin, stretching, splinting, and orthosis was reportedly useful in improving contracture of the spastic children.<sup>21-23</sup>

Ultrasound therapy,<sup>17</sup> massages, and stretching of muscles may decrease spasticity, promote muscle relaxation, and prevent contractures<sup>20</sup> with the effect lasting from 30 minutes to 2 hours. For a longer duration of effect, the stretch on the muscle should be maintained for several hours every day. Ultrasound therapy in a continuous

mode applied together with the stretching exercise, can improve extensibility of the tendon and muscles as well as provide relaxation in the spastic muscles<sup>24</sup> facilitated by 24-hour postural management at home.<sup>20</sup> The strategies include low-load active stretching which increases the range of movement, and passive stretching with positioning and use of orthoses.<sup>19,20</sup> Prolonged stretch of the spastic muscles can decrease the sensitivity of the stretch reflex and the brain stem reflexes that trigger spasticity.<sup>20</sup> A review reported that 24-hour postural management reduced the effects of postural asymmetries and prevented the development of deformities in individuals with immobilization.<sup>25</sup>

Children with brain lesions may be unable to modify and control body positions. Therefore, proper comfortable positioning and postural support to keep the body in the right posture while sitting or lying on the bed should be considered. The family should be taught about these positions to assist the child most of the time at home.<sup>20</sup> In this case, the mother was educated to place the patient in a comfortable symmetrical position, with the head, trunk, and limbs supported and fixed to the optimal position. Ankle foot and wrist hand orthoses were also prescribed. Assistive devices such as wedges or pillows were provided to ensure proper positioning at home.<sup>19,20</sup> After four weeks of rehabilitation program, the pain intensity decreased from severe to moderate, the posture asymmetry was improved, and the mother could carry out positioning more easily.

## CONCLUSION

In conclusion, pain is a complex condition in children with meningoencephalitis, which needs

evaluation and management. The multiple sources can be assessed using the ICF framework, while the management of pain should be multidisciplinary and multi-modality. Furthermore, it is important to engage the family in the treatment, as 24-hour postural management at home is an important part of rehabilitation management. This case report underscores the need for further study into pain evaluation and treatment for children with brain lesions.

## PATIENT CONSENT

Consent had been given by the mother of the patient to publish information in this journal.

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