

Clinical case

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Severe rigid hip flexion-abduction contracture in cerebral palsy: a case report and review of the literature

*Contractura rígida grave de flexión-abducción de cadera en parálisis cerebral: reporte de un caso y revisión de la literatura*Ramírez-Barragán A,* Galán-Olleros M,*[‡] Egea-Gámez RM,* Palazón-Quevedo A,* Martínez-Caballero I*

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ABSTRACT. Introduction: severe, rigid hip abduction deformity in individuals with cerebral palsy (CP) is an exceptionally uncommon condition. This posture hinders the positioning in the wheelchair and the completion of basic activities of daily living (ADL). Addressing such severe deformities can be quite challenging. **Material and methods:** a 14-year-old male, with spastic-dystonic quadriplegic CP, developed rigid and severe flexion-abduction contractures in both hips, characterized by 90 degrees of flexion and 100 degrees of abduction. These contractures severely impeded his ability to comfortably use a wheelchair and even pass through doorways. Performing basic ADLs became a significant challenge for both the patient and his caregivers. **Results:** the treatment approach involved a two-stage surgical procedure, one for each hip, with a two-month interval between them. An extensive release of the fascia latae, gluteus maximus, external rotators, and hip flexors; in combination with a proximal femur osteotomy were performed. To maintain the corrections achieved, long-leg casts connected with two bars were employed, followed by orthotic support and physiotherapy. Following the procedure, lower limb adduction was achieved, and the patient and caregivers were highly satisfied, as ADLs and basic caregiving had

RESUMEN. Introducción: la deformidad severa y rígida en abducción de cadera en individuos con parálisis cerebral (PC) es una condición infrecuente. Esta postura dificulta el posicionamiento en la silla de ruedas y la realización de actividades básicas de la vida diaria (AVD). El tratamiento de estas deformidades tan severas puede ser todo un reto. **Material y métodos:** varón de 14 años, con PC tetrapléjica espástica-distónica, que desarrolló contracturas rígidas y severas de flexión-abducción en ambas caderas, caracterizadas por 90 grados de flexión y 100 grados de abducción. Estas contracturas impedían gravemente su capacidad para utilizar cómodamente una silla de ruedas e incluso pasar por las puertas. La realización de actividades básicas de la vida diaria se convirtió en un reto importante tanto para el paciente como para sus cuidadores. **Resultados:** el tratamiento consistió en una intervención quirúrgica en dos fases, una para cada cadera, con un intervalo de dos meses entre ellas. Se realizó una amplia liberación de la fascia lata, el glúteo mayor, los rotadores externos y los flexores de la cadera; en combinación con una osteotomía proximal del fémur. Para mantener las correcciones conseguidas, se emplearon escayolas de pierna larga conectadas con dos barras, seguidas de soporte ortésico y fisioterapia. Tras la intervención, se consiguió la aducción de los miembros inferiores y el paciente y los cuidadores se

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been greatly facilitated. **Conclusions:** while the available literature on the management of severe rigid abduction hip contractures in non-ambulatory CP patients is limited, and treatment options are often complex, the present case underscores the effectiveness of a comprehensive approach involving soft tissue release and bone surgery. Achieving a more favorable wheelchair positioning and facilitating basic ADLs and care represents a significant success for patients and families.

Keywords: cerebral palsy, hip contracture, surgical release, activities of daily living, health-related quality of life.

mostraron muy satisfechos, ya que se habían facilitado en gran medida las AVD y los cuidados básicos. **Conclusiones:** aunque la bibliografía disponible sobre el tratamiento de las contracturas rígidas graves de la cadera en abducción en pacientes no deambulantes con PC es limitada, y las opciones de tratamiento suelen ser complejas, el presente caso subraya la eficacia de un enfoque integral que incluye la liberación de los tejidos blandos y la cirugía ósea. Conseguir una posición más favorable en la silla de ruedas y facilitar las AVD básicas y los cuidados representa un éxito significativo para los pacientes y sus familias.

Palabras clave: parálisis cerebral, contractura de cadera, liberación quirúrgica, actividades de la vida diaria, calidad de vida relacionada con la salud.

Introduction

Severe, rigid flexion-abduction hip contractures represent a rare occurrence in cerebral palsy (CP) patients.¹ This specific hip positioning, characterized by flexion and abduction, is more prevalent in children with generalized hypotonia and is less commonly observed in purely spastic CP cases. Typically, spasticity affecting the hip adductors and flexors is more common, especially in non-ambulant CP patients.²

Hip abductor muscle contractures have been documented in patients with conditions such as poliomyelitis, gluteal fibrosis resulting from infection, a history of intramuscular injections, and fibrosis of unknown origin in the gluteus maximus. In these instances, muscular fibrosis restricts hip mobility, causing contractures in the abductor and external rotator muscles. The preferred treatment is surgical, involving Z-lengthening of the fibrous bands.^{3,4}

In CP, this deformity can develop as a result of flexor/adductor extensive release combined with anterior branch obturator neurectomy,⁵ potentially leading to pain and hampering the child's ability to sit, stand, or transfer. The initial approach is non-surgical, encompassing physiotherapy, orthotic intervention, proper positioning, and targeted spasticity management using Botulinum toxin type A injections to prevent or mitigate joint deformities. Musculotendinous lengthening or orthopedic procedures may be required in more severe cases.^{6,7}

Spasticity involving the hip flexors and abductors poses a considerable challenge for children with severe motor impairments associated with CP. Despite the substantial impact of this severe deformity, there is limited research in the existing literature regarding its treatment in CP patients.^{1,3} This report aims to delineate the management of a severe hyperabduction contracture in an adolescent with CP while providing an overview of the available literature on the subject.

Case report

A 14-year-old boy with spastic-dystonic quadriplegic CP, classified as gross motor function classification

system (GMFCS) level V, presented to our center. He had mild intellectual disability but could communicate with his caregivers and had no respiratory or gastrointestinal issues. Born in Equatorial Guinea, he had been consistently maintained in a flexion-abduction position since childhood to prevent hip dislocation. He did not receive surgical or orthotic treatment, nor physiotherapy during his early years, remaining chair-bound without ever having the opportunity to stand, even with the support of a standing frame.

Upon physical examination, the patient exhibited fixed flexion, hyperabduction, and external rotation of the hips, with limited lower limb adduction capability. His hips were flexed to 90° and abducted to 100° in a supine position. Bilateral knee flexion contractures were observed, partially reducible to 60° (*Figure 1A*). Additionally, he displayed rigid planus valgus feet and wrist deformities. No spinal deformities were detected (*Figure 1B*).

With the onset of adolescence, the patient's caregivers faced increasing challenges in his daily care, including limited wheelchair tolerance and difficulty passing through doorways. Despite the patient's lack of hip pain, a decision was made to pursue surgical intervention with the primary goal of enhancing his activities of daily living (ADLs) and overall sitting comfort.

The surgical procedure was divided into two stages, starting with the right hip and followed by the left hip two months later. The patient was placed in lateral decubitus after general anesthesia and placement of an epidural catheter (*Figure 2A and B*). We first performed percutaneous hamstring tenotomy, proximal tenotomy of the fascia latae, myotomy of the posterior gluteus medius proximal to the greater trochanter were performed. The gluteus minimus and short external rotators were also detached from their insertions on the greater trochanter. A posterior capsulotomy was also needed to address contracture. Subsequently, anterior incisions were made to release the sartorius and rectus anterior. Through a third longitudinal incision made on the distal lateral thigh, we resected 2 cm of the iliotibial band. This was followed by a varus, extension, derotational and 2 cm shortening proximal femoral osteotomy.

The locking cannulated blade plate system was used to compress the osteotomy, achieving adduction, extension, and a derotational effect on the lower extremity. At that time, an iliopsoas tenotomy was performed at its insertion into the lesser trochanter. Surgical wounds were closed in the usual manner, and a drain was left in place for 24 hours. The patient was immobilized in long-leg casts, with the right lower limb in adduction and maximum hip-knee extension, keeping the two limbs connected by two bars (*Figure 2C*). Three weeks later, the immobilization device was replaced with a thigh orthosis in the same position.

The left hip underwent a similar procedure two months later. After surgery, the patient wore a thigh orthosis with the hips in 20° abduction for approximately 18 hours per

day for three months and thereafter at nights throughout the first year (*Figure 2D*). The patient also underwent physiotherapy to improve hip, knee, and ankle mobility, as well as the upper limbs. Six months postoperative, lower limb alignment had significantly improved, his passive hip range of movement had improved bilaterally (*Table 1*), and the patient could sit and lie comfortably.

Three years after the procedure, the patient maintained excellent correction of hip contractures and lower limb alignment. He achieved a proper wheelchair position, facilitated passage through doorways, and alternated between lateral and supine sleeping positions (*Figure 3*). Caregivers found the handling of the patient significantly improved and reported

Figure 1:

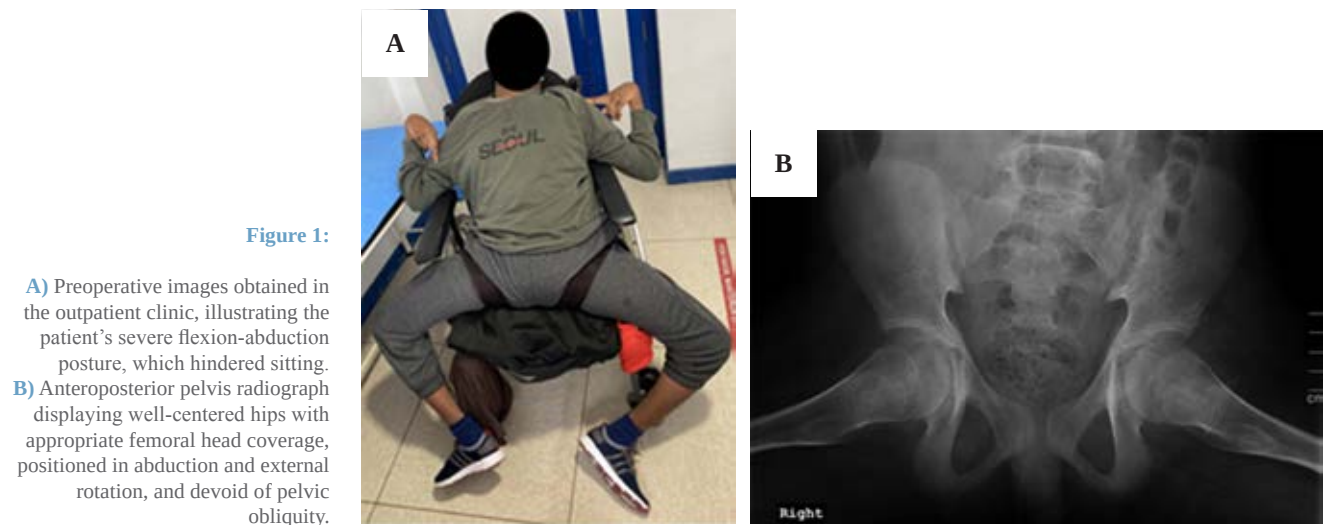


Figure 2:

A) Image of the patient lying supine on the operating table. **B)** Patient's positioning in lateral decubitus for right hip surgery, revealing the pronounced deformities of the hips, knees, and feet. **C)** Postoperative immobilization utilizing long-leg casts connected by two bars to maintain the corrected position. **D)** Position with thigh orthoses, retaining the hips in 20° abduction.

increased satisfaction, as reflected in the caregiver priorities and child health index of life with disabilities (CPCHILD) questionnaire⁸ score, which rose from 44.34 to 61.32 (*Table 1*).

Discussion

CP is a condition stemming from brain damage occurring before or during birth or in early childhood, characterized

by progressive musculoskeletal changes.⁹ Children with spastic CP often experience evolving muscle contractures, bony deformities, and joint stiffness, with clinical presentations varying between individuals. Hip adductor and flexor spasticity are the most common culprits, leading to fixed contractures, muscle imbalances, and progressive hip displacement.^{2,10,11} Among non-ambulatory patients, the «windblown hip» deformity is prevalent,^{12,13} where one hip assumes an abducted and externally rotated position while the contralateral hip is adducted and internally rotated. This positioning predisposes the patient to hip dislocation, both anteriorly in abduction and posteriorly in adduction.¹⁴

Severe hip flexion-abduction contractures, as observed in this case, are a rare but significantly challenging deformity in CP. Patients with such severe contractures encounter substantial difficulties in sitting in a wheelchair, navigating doorways during adolescence, and assuming various lying positions.^{1,11,15}

From a biomechanical perspective, hip abduction deformities can arise from contractures within different muscle groups. The gluteus maximus, typically a hip extensor in the standing position, contributes to hip abduction while sitting. Contracture in this muscle leads to a fixed abduction of the lower limbs when seated.^{16,17} Tightness in the iliotibial band may also contribute to the deformity, resulting in hip flexion, abduction, and external rotation.¹⁸ A review of the literature did not reveal a direct association between flexion-abduction hip positioning and hip dislocation,¹⁹ as confirmed in our patient. However, it may be related to the development of acetabular protrusion due to increased medial acetabular pressure, although no radiological evidence of this was observed in our case.

Table 1: Hip range of motion of the patient (right/left) and CPGHILD score.

	Preoperative	Postoperative
Range of motion (degrees)		
Flexion	100°/100°	100°/100°
Extension	-70°/-60°	-15°/-10°
Abduction	100°/100°	50°/40°
Adduction	-80°/-90°	-10°/-15°
Internal rotation	10°/15°	30°/30°
External rotation	70°/60°	40°/40°
CPCHILD score		
1. Personal care & activities of daily living	34.57	50.62
2. Positioning, transferring, and mobility	8.33	41.67
3. Comfort & emotions	71.43	77.78
4. Communication & social interaction	57.14	71.43
5. Health	66.67	73.33
6. Overall quality of life	20.00	60.00
Total score	44.34	61.32

CPCHILD = caregiver priorities and child health index of life with disabilities.

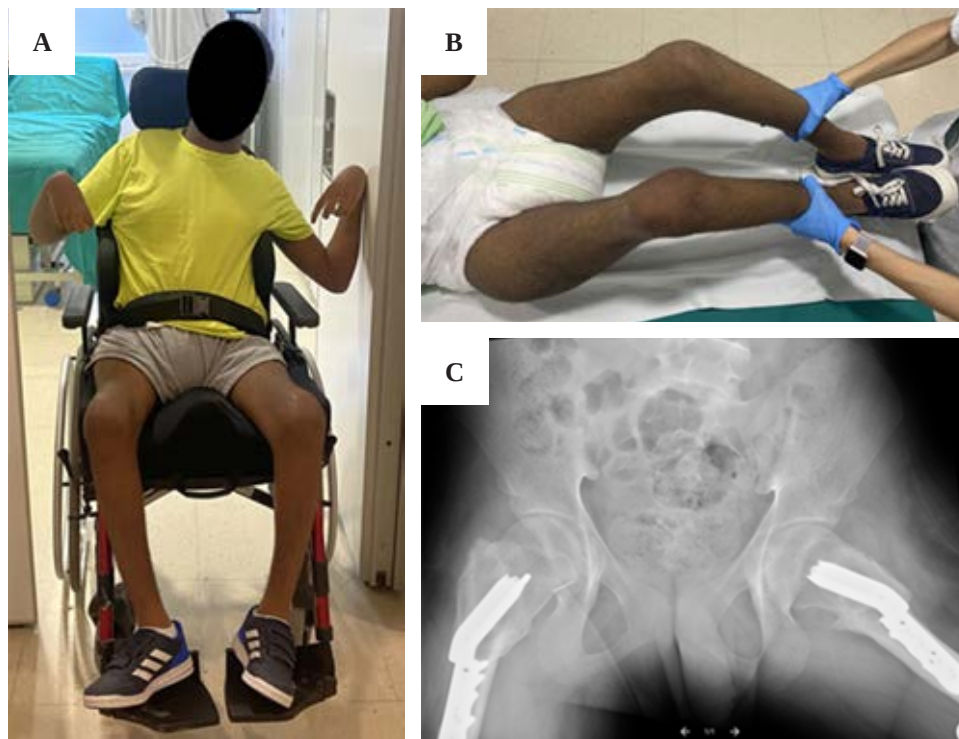


Figure 3:

A) Patient comfortably seated in a wheelchair, benefitting from effective positioning. **B)** Patient in the supine position, displaying the hips in a less severe degree of abduction. **C)** Postoperative radiograph, demonstrating both extremities closer to the midline with well-centered hips.

The approach to addressing fixed flexion-abduction hip deformities in CP hinges on the severity of the deformity.^{5,20} When passive adduction of the hip to the midline can be achieved, the preferred treatment involves muscle-tendon lengthening. Correction of the deformity entails myotomy of the posterior gluteus medius just above the greater trochanter, sectioning the gluteus maximus just proximal to its insertion on the femur, and detaching all short external rotators, including the piriformis. In cases where the iliotibial band is significantly contracted, release of both its proximal and distal segments (the Ober-Yount procedure) can enhance the positioning of the hips and knees. Furthermore, hamstring lengthening is frequently necessary. For patients with more severe and rigid flexion-abduction contractures, particularly in adolescents, such as the case we presented, a concomitant proximal femoral osteotomy to reposition the lower extremity is often indispensable.⁵

Casting and orthotic treatment, followed by subsequent physiotherapy, play pivotal roles in achieving favorable outcomes post-surgery, as exemplified in our case. We recommend initiating physiotherapy as early as possible to mobilize the various joints and facilitate wheelchair placement, which promotes hip adduction.

Conclusion

In conclusion, despite the limited literature on managing severe rigid hip flexion-abduction contractures in non-ambulatory CP patients, our case underscores the effectiveness of treatment through soft-tissue release and bone surgery, followed by casting, orthotic intervention, and physiotherapy. The attainment of optimal wheelchair positioning and improvements in ADL and basic care signifies a successful outcome for both patients and caregivers.

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Reporting guidelines: authors have adhered to the recommendations for the accurate reporting of information in case reports that resulted in the CARE (CASE Report) statement and checklist.