Case Report

Functioning of a child with bilateral cerebral palsy submitted to multilevel surgery: case report

Funcionalidade de uma criança com paralisia cerebral bilateral submetida a cirurgia multinível: relato de caso

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ABSTRACT | INTRODUCTION: Interventions in different joints may be necessary to correct crouch gait and other musculoskeletal changes that occur as time passes for Cerebral Palsy (CP) children. Multilevel surgery reduces the number of hospitalizations, contributes to the prevention of secondary disabilities, and improves the ambulation ability of children with diplegia. **OBJECTIVE:** Document the changes in mobility outcomes of a bilateral CP child, type diplegia, child after lower limbs multilevel surgery in the Brazilian context. **MATERIAL AND METHODS:** The participant was an eight-year-old girl, Gross Motor Function Classification System level III. The mother signed the informed consent form. The Gross Motor Function Measure–66 (GMFM-66) was performed one day before surgery and one, three, six, and twelve months after surgery. Additional information was obtained through the electronic system of the hospital to complete the child's evolution records. **RESULTS:** The GMFM-66 total score was: 49.6 pre-operative (CI95%: 47.3-51.9); 42.8 after one month (CI95%: 40.7-45.0); 49.9 after three months (CI95%: 47.6-52.1); 52.6 after six months (CI95%: 50.2-55.0) and 56.9 after one year (CI95%: 54.6-59.2), increasing after 6 months of surgery (*p<0.05). The participant presented satisfactory adherence to physiotherapy. **CONCLUSION:** This study describes the case of a Brazilian child with CP using the public health system. The impact of multilevel surgery was predominant in mobility, with worsening of capacity soon after surgery and progressive improvement over the months. Factors that may have contributed to our results were adherence, frequency, contextual factors.

KEYWORDS: Cerebral Palsy. Multilevel Surgery. Functioning. Case report. Physiotherapy.

RESUMO | INTRODUÇÃO: Intervenções em diferentes articulações podem ser necessárias para corrigir a marcha agachada e outras alterações musculoesqueléticas que ocorrem com o passar do tempo em crianças com Paralisia Cerebral (PC). A cirurgia multinível reduz o número de hospitalizações, contribui para a prevenção de deficiências secundárias e melhora a capacidade de locomoção de crianças com diplegia. **OBJETIVO:** Documentar as mudanças na mobilidade de uma criança com PC bilateral, do tipo diplégica, após cirurgia multinível de membros inferiores na realidade brasileira. **MATERIAIS E MÉTODOS:** A participante foi uma menina de 8 anos, Gross Motor Function Classification System nível III. A mãe assinou o termo de consentimento livre e esclarecido. O Gross Motor Function Measure-66 (GMFM-66) foi realizado um dia antes da cirurgia e um, três, seis e doze meses após a cirurgia. Para completar os registros da evolução da criança, foram obtidas informações adicionais por meio do sistema eletrônico do hospital. **RESULTADOS:** O escore total do GMFM-66 foi: 49,6 pré-operatório (IC95%: 47,3-51,9); 42,8 após um mês (IC95%: 40,7-45,0); 49,9 após três meses (IC95%: 47,6-52,1); 52,6 após seis meses (IC95%: 50,2-55,0) e 56,9 após um ano (IC95%: 54,6-59,2), aumentando após seis meses da cirurgia (*p<0,05). A participante apresentou adesão satisfatória à fisioterapia. **CONCLUSÃO:** Estudo de caso de criança brasileira com PC, usando o sistema único de saúde. O impacto da cirurgia multinível foi predominante na mobilidade, com piora da capacidade logo após a cirurgia e melhora progressiva ao longo dos meses. Os fatores que podem ter contribuído para nossos resultados foram adesão, frequência, fatores contextuais.

PALAVRAS-CHAVE: Paralisia cerebral. Cirurgia multinível. Funcionalidade. Relato de caso. Fisioterapia.

Submitted 05/26/2021, Accepted 07/19/2021, Published 08/02/2021
J. Physiother. Res., Salvador, 2021 November;11(4):xx-xx
http://dx.doi.org/10.17267/2238-2704rpf.v11i4.3851 | ISSN: 2238-2704
Designated editors: Hércules Leite, Katia Sá

How to cite this article: Rosa RM, Valenzuela EJ, Defilipo EC, Chagas PSC. Functioning of a child with bilateral cerebral palsy submitted to multilevel surgery: case report. J Physiother Res. 2021;11(4):xx-xx. http://dx.doi.org/10.17267/2238-2704rpf.v11i4.3851

Introduction

The health status of children with Cerebral Palsy (CP) changes over the years under the influence of personal factors such as age and environmental factors such as interventions.[1-3] Time does not contribute positively to improve performance in the case of children with diplegia.[1] Worsening in the gait pattern sometimes leads to using gait aids such as walkers and the development of modified gait patterns, especially the crouch gait.[4]

Interventions in different joints may be necessary to correct crouch gait and other musculoskeletal changes that occur as time passes. Considered the standard of care for disability interventions, multilevel surgery reduces the number of hospitalizations and rehabilitation time, contributes to the prevention of secondary disabilities, and improves the ambulation ability of children with diplegia.[5]

A strong relationship has been reported between the favorable prognosis of surgery on functioning and the child's age when it is performed: the better the functioning; the earlier surgery is necessary to optimize their motor development.[3,6] In addition, studies show the beneficial effect of multilevel surgery on the CP child.[1,7-10] In Brazil, however, there is no guarantee that children will receive evidence-based interventions in time due to barriers to implementation in systematized clinical practice within the Public Health System (SUS), as there is still a gap between research and clinical practice.[11]

Children with CP with a lower socioeconomic profile are predominantly dependent on the SUS, in the Brazilian context, and require continuous monitoring of health professionals and interventions for many years of life.[11,12] Considering that time is crucial for children with CP who require surgery and can often be decisive for their long-term positive outcomes[2,13], standardized assessments, with clinical significance interpretation and timely documentation of the outcomes can help us validate appropriate physical therapy care in public clinical practice in Brazil.

The International Classification of Functioning, Disability, and Health (ICF) defines Functioning as a term covering body functions, activities, and participation.[13] Some aspects of Functioning cannot be observed directly; they must be inferred through standardized tests. For other aspects, such as contextual factors, self-reported data may be considered reliable and meaningful.[13]

This study aimed to document and describe the Functioning of a bilateral CP child, type diplegia, using a standardized instrument (GMFM-66) to document the activity (mobility) in the pre and postoperative period of multilevel surgery for the lower extremities and to document the contextual factors.

Case description

Type of Study and Ethical Aspects

Observational, retrospective, case report study, approved by the Research Ethics Committee of the University Hospital of Universidade Federal de Juiz de Fora (CAAE: 09581119.1.00005133). The mother signed the Informed Consent Form and authorization to use images.

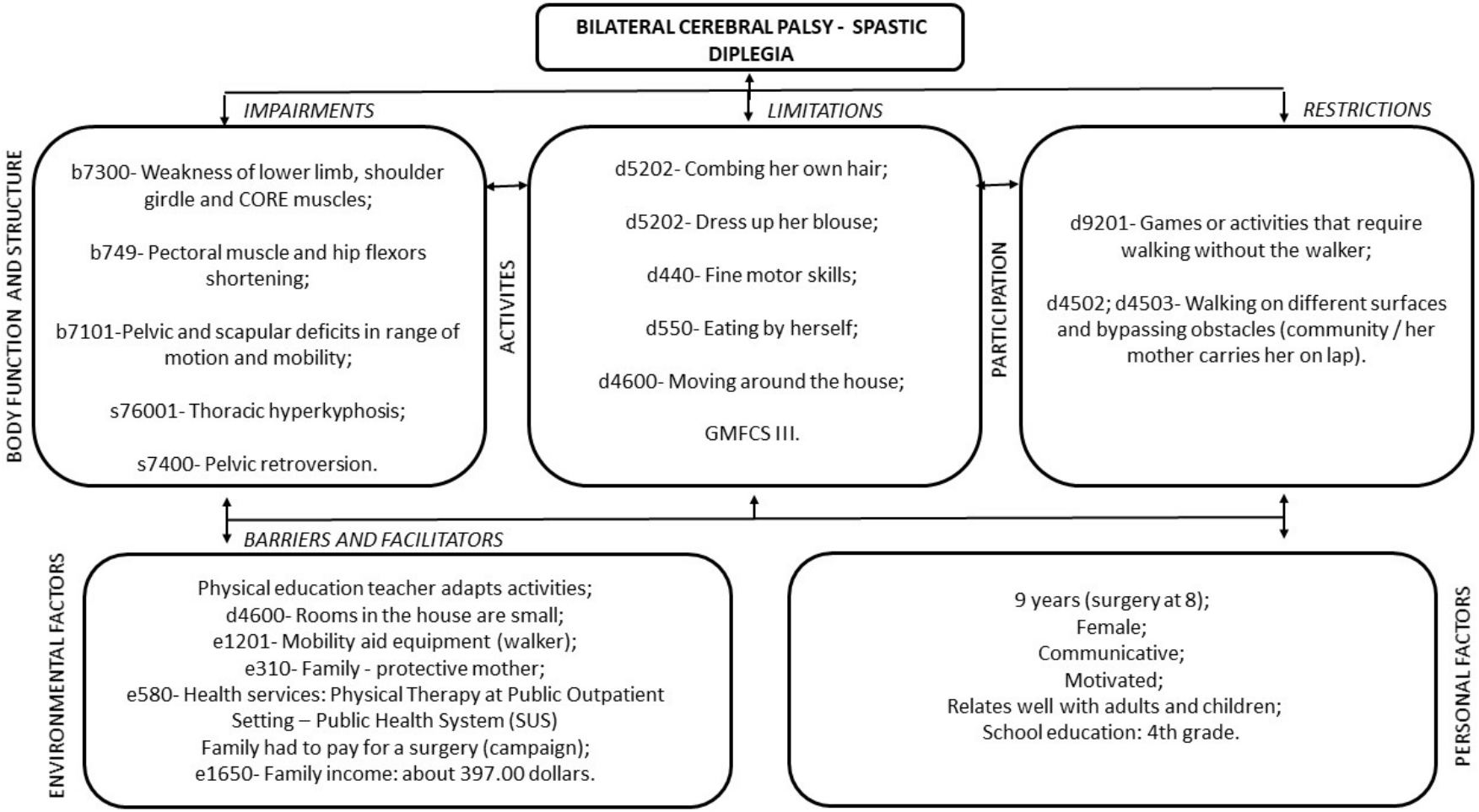
Participant

An eight-year-old female child with bilateral CP and Gross Motor Function Classification System (GMFCS) level III, cared for by the physiotherapy clinic of the University Hospital. She lives with her mother, stepfather, and little brother. Family income is about 397.00 dollars. They use primarily the public health system including physiotherapy treatment. As a GMFCS III, the walker is necessary, and her home environment is a barrier to her adequate mobility: rooms are small, and she cannot use the walker inside the house, so she crawls. The access to enter the house is through stairs, with no handrail, and she cannot climb the stairs on her own. Instead, she needs to be carried on. She is a communicative and friendly child presenting great interaction with adults and children. She lives in the city of Juiz de Fora, Minas Gerais. She was in the 4th grade of elementary school, assisted by a support teacher.

Before the surgery

In figure 1, we present the participant according to the International Classification of Functioning (ICF) biopsychosocial model, based on the registries on the Hospital's electronic medical record.

Figure 1. Descriptive table of the participant's characteristics according to the International Classification of Functioning, Disability and Health (ICF) biopsychosocial model



Interventions

She underwent the multilevel surgical procedure for the lower extremities in February 2018, at the age of eight. The surgical intervention consisted of incomplete open release of adductor longus and gracilis, preserving anterior branch obturator nerve and fascia lengthening of the psoas for hip flexion deformity; equinus deformity correction by lengthening of the gastrocnemius fascia and soleus, lengthening of the medial hamstrings to correct flexed knee. The goals of these procedures were to improve walking abilities, preserve joint mobility, and reduce the risk of pain as she grows older. The surgery took place in a private health service, with money raised in a campaign carried out by her mother.

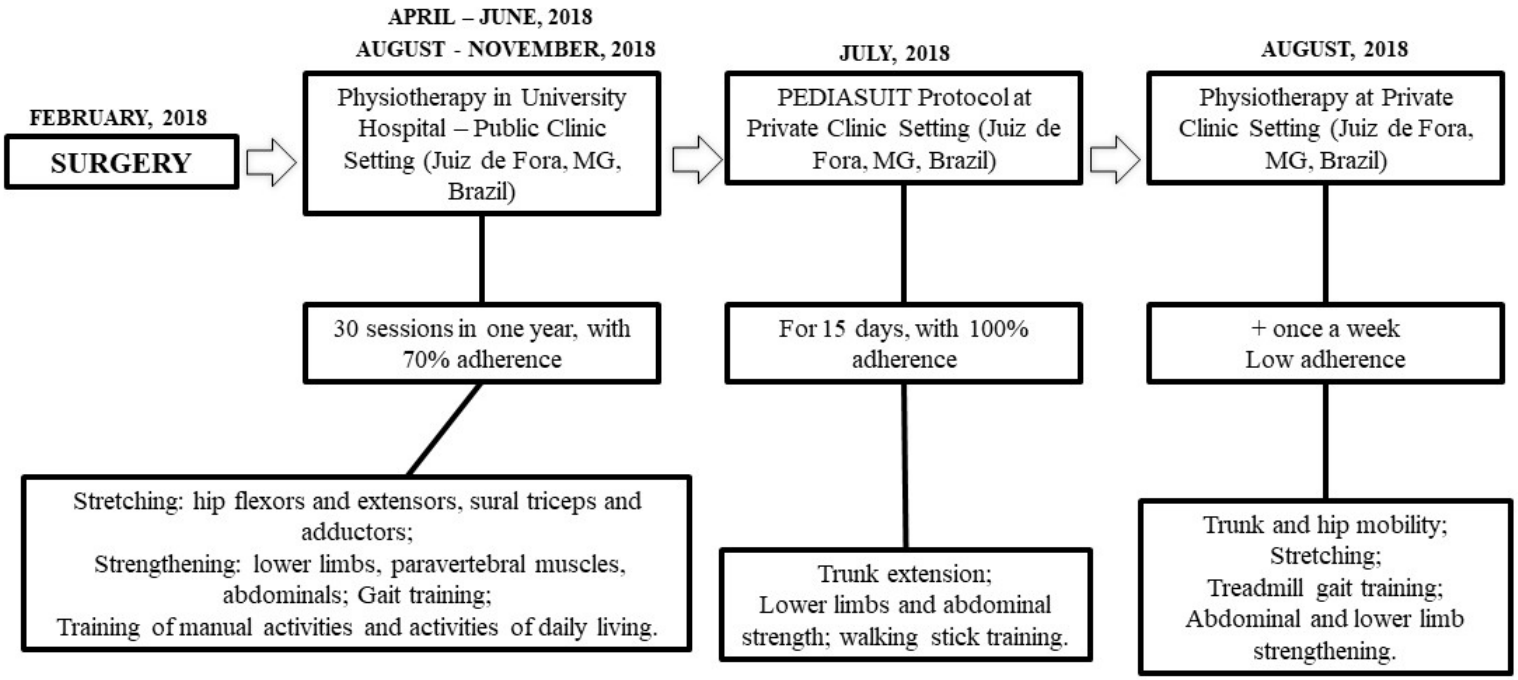
According to the electronic medical record of the Hospital, physical therapy evaluation after surgery showed: weakness of lower limbs, shoulder girdle, and CORE muscles; pectoral muscles and hip flexors shortening; pelvic and scapular deficits in range of motion and mobility; thoracic hyperkyphosis and pelvic retroversion. These dysfunctions influence limitations in activities such as combing her own hair, dress up her blouse, eating by herself, and walking without an assistive device. In addition, she presented restrictions to participate in games and activities at school and walking in the community because of the obstacles and different surfaces, resulting in the need to be carried by her mother.

Two months after surgery (April 2018), the physical therapy interventions began aiming to stretch and strengthen lower limbs musculature, gait training with a walker, and training of manual activities of daily living at a public hospital. After the first three months of physical therapy, she regained the activity profile she showed before surgery. At this point, we decided that the participant would benefit from more intensive therapy, especially with the focus on muscle strengthening, postural alignment, and self-confidence.

In July, the PEDIASUIT® Protocol at the private clinic setting started, during two weeks of the winter holiday, five days a week, for three hours, totalling 30 hours of intensive intervention. It is an intensive intervention using a therapeutic suit, where it is possible to perform the activities against the resistance given by the elastics, increasing proprioceptive information and postural realignment associated with the specific goals of therapy.[14] After this protocol, the participant went back to a frequency of 3 times per week of conventional physical therapy treatment to maintain the gained skills and train new acquisitions. During all the treatment care, the child used an anterior walker and a pair of articulated ankle-foot orthoses custom-made.

Figure 2 shows details of adherence and the timeline of therapies she underwent.

Figure 2. Locations, frequency of sessions and physical therapy interventions performed during the one-year postoperative period



Measures and outcome

Outcome Measures

To describe the participant's level of functioning, we used the Gross Motor Function Classification System (GMFCS).[15] Another instrument used in our assessment was the Gross Motor Function Measure[15] to assess the activity (mobility).

We used all dimensions of GMFM-66, and for the analysis of the total scores, we used the GMAE-2 software. We calculated Expected Natural Evolution (ENE) from the participant's age, according to the evaluation scores and the time interval between them in the www.gmfmer.ca[16] (Table 1).

In addition to documenting the evolution of the participant in this study, we also consulted the health records of pre and postoperative evaluations and physiotherapy care performed over one year (including frequency and adherence, number of sessions and activities performed), data obtained through the electronic medical record of the Hospital.

Data Analysis

We used a descriptive analysis of the case, reporting all treatment stages and health status evolution according to GMFM-66. In addition, the Gross Motor Function Measure Evolution Ratio (GMFM-ER) was used to calculate the expected natural evolution of this child's score using age, initial GMFM-66 score, and the time interval between assessments.[15] For the statistical interpretation, differences between the months of intervention, the data were analyzed based on the difference between the total score on the GMFM-66 surpassing the higher value of the confidence interval of 95% ($p<0.05$).

Results

Table 1 shows the total score on the GMFM-66 per month evaluated, the confidence interval of 95%, and the changes in points obtained in each evaluation according to the preoperative GMFM-66. Also, this table shows the expected change according to the ENE and the actual change observed in the GMFMER. The child began to show significant changes from the 6 months forward after the surgery ($p<0.05$).

Table 1. Gross Motor Function Measure test score (GMFM-66) and percentage changes obtained over one year postoperatively

ASSESSMENT/ MONTH	SCORE OBTAINED (MEAN/ CONFIDENCE INTERVAL)	CHANGES IN POINTS RELATED TO PREOPERATIVE (GMFM score change)	PERCENTAGE CHANGES FROM PREOPERATIVE	EXPECTED CHANGE IN POINTS * (ENE)	GMFMER (GMFM score change/ ENE)
Preoperative	49.6 (47.3-51.9)	-	-	-	-
1 month PO	42.8 (40.7-45.0)	-6.8[a]	13.71%[a]	0.02	-340.00
3 months PO	49.9 (47.6-52.1)	+0.3[c]	+0.60%[c]	0.06	2.50
6 months PO	52.6 (50.2-55.0)	+3.0[b,c]	+6.04%[b,c]	0.12	25.00
9 months PO	55.2 (52.7-57.6)	+5.6[b,c]	+11.29%[b,c]	0.18	31.11
12 months PO	56.9 (54.6-59.2)	+7.3[b]	+23.32%[b]	0.22	33.18

Legend: a: significant decrease in the value obtained in the test compared to the preoperative; b: significant increase in the value obtained in the test compared to the preoperative; c: significant increase in the value obtained in the test compared to the immediately previous evaluation; ENE: Expected Natural Evolution; GMFM change score: changes in preoperative points; GMFMER (Gross Motor Function Measure Evolution Ratio): Relationship between observed point change and expected natural evolution; PO: postoperatively. * GMFM ratio[16]

Implications

The present study is relevant because it shows the case of a Brazilian child with bilateral CP, a user of the public health system, with positive results in her mobility after surgical intervention. Contextual factors may have contributed to the benefits found: family dedication, patient motivation, and the interventions, such as the surgery at the ideal moment of musculoskeletal growth and the rehabilitation program she underwent. Our study reports a percentage gain in GMFM-66 greater than 20% after 12 months of surgery, exceeding previous results in the literature. For this result, we consider it important to mention the contextual factors of the case presented, such as child motivation and environmental support and the intensive protocol associated with conventional PT.

For the future, there is a need for a larger study investigating the contextual factors in a larger population in Brazil in order to be able to broaden interpretation in different personal and environmental contexts, enabling therapists to target their interventions better. We also consider important further studies analyzing intensive rehabilitation after surgery, such as the PEDIASUIT protocol. Thus, it may be possible to interpret better how this kind of intervention contributes to greater gains in the functioning of these children after the surgical procedure. In addition, further studies using the GMFM-ER would allow for more discussions and comparison of results and interpretation of outcomes.

Conclusion

Multilevel surgery improves gait in children with diplegia and improves GMFM-66 scores about 2% after one year when associated with conventional PT. Our study reports a percentage gain in GMFM-66 greater than 20% after 12 months of surgery, exceeding previous results in the literature. We consider it important to analyze the contextual factors and the intensive protocol associated with conventional PT to achieve such results. However, it is essential to analyze the variables of our study in a larger population to broaden interpretation.

Acknowledgments

To the Physical Therapist Natalia Trindade of the Núcleo Integrado de Desenvolvimento for providing PEDIASUIT-protocol and to doctor Jordana Caifa for the surgical procedure in the patient.

Author contributions

Rosa RM planned the study, conducted the evaluations, wrote the manuscript, and analyzed the data. Valenzuela EJ participated in the planning of the study, reviewed the manuscript and the data analysis. Defilipo EC reviewed the manuscript and the data analysis. Chagas PSC planned the study, conducted the evaluations, wrote the manuscript, and analyzed the data. All authors have had full access to study data and support a publication, read a final version of this manuscript, and agree to be submitted to this journal for possible publication.

Competing interests

No financial, legal, or political competing interests with third parties (government, commercial, private foundation, etc.) were disclosed for any aspect of the submitted work (including but not limited to grants, data monitoring board, study design, manuscript preparation, statistical analysis, etc.).

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